The University Of Sheffield.

The relationship between Stigma and Self-Efficacy in individuals with epilepsy or nonepileptic attack disorder

By:

Lewis Hanney

A thesis submitted in partial fulfilment of the requirements for the degree of

Doctor of Clinical Psychology

The University of Sheffield

Faculty of Science

Clinical Psychology Unit, Department of Psychology

Submission Date: November 2019

The University Of Sheffield.

Access To Thesis.

This form must be completed and returned to Research Services prior to the award of your degree

SECTION 1: STUDENT DETAILS			
Family Name	Hanney	First Name	Lewis
Registration Number	160124536	Department	Psychology
Thesis Title	The relationship between Stigma and Self-Efficacy in individuals with epilepsy or nonepileptic attack disorder		

SECTION 2: THESIS SUBMISSION DETAILS – PLEASE SELECT ONE OF THE FOLLOWING OPTIONS	
☐	I am submitting in print format only for deposit in the University Library **(Note: this option only applies to students who initially registered prior to 2008)**
☐	I am submitting an eThesis only to the White Rose eTheses Online server. I confirm that the eThesis is a complete version of my thesis and no content has been removed
☒	I am submitting an eThesis to the White Rose eTheses Online server and also submitting in print format because I have removed some content from my eThesis

SECTION 3: EMBARGO DETAILS – PLEASE SELECT FROM THE FOLLOWING OPTIONS

Requests for embargoes that exceed 12 months will require Faculty approval. If you wish to request a longer embargo, please also complete the extended embargo request form, available at: www.sheffield.ac.uk/rs/code/embargoes and return it to pgr-enquiries@sheffield.ac.uk

If no boxes are ticked, you will be deemed to have consented to your thesis being made available immediately without any embargo.

Should the thesis be embargoed? If 'Yes', please specify the length of embargo requested (in years)					
	Print Thesis (where applicable)	☐ No	☒ Yes	1	Years
	eThesis	☐ No	☒ Yes	1	Years

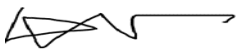
Reason for the embargo (please select from the following options):

☐	Third party copyright	☐	Commercial confidentiality
☐	Contains personal data	☐	Could prejudice national security
☐	Could endanger health and safety	☐	Exempt under another category listed in the FOI Act 2000
☒	Planned publication	☐	Other (please specify)

SECTION 4: COPYRIGHT LICENCE OPTIONS – PLEASE SELECT ONE OF THE FOLLOWING

This thesis is protected by the Copyright Design and Patents Act 1988. No reproduction is permitted without consent of the author. It is recommended that you make your thesis available using a Creative Commons Licence https://creativecommons.org/share-your-work/. This Licence protects you as the author of the work and also clarifies the uses that others may make of your work.

☒	Creative Commons Attribution-Non-Commercial-No-derivatives **(recommended)**	☐	Creative Commons Attribution-Non-Commercial
☐	Creative Commons Attribution	☐	Creative Commons Attribution-No-derivative-Works
☐	Creative Commons Attribution-Non-Commercial-Share Alike	☐	Other/Do not apply a Licence

SECTION 5: THESIS DEPOSIT AGREEMENT - STUDENT		
1. I, the author, confirm that the Thesis is my own work, and that where materials owned by a third party have been used, copyright clearance has been obtained. I am aware of the University's *Guidance on the Use of Unfair Means* (www.sheffield.ac.uk/ssid/exams/plagiarism).		
2. I confirm that all copies of the Thesis submitted to the University, whether in print or electronic format, are identical in content and correspond with the version of the Thesis upon which the examiners based their recommendation for the award of the degree (unless edited as indicated above).		
3. I agree to the named Thesis being made available in accordance with the conditions specified above.		
4. I give permission to the University of Sheffield to reproduce the print Thesis (where applicable) in digital format, in whole or part, in order to supply single copies for the purpose of research or private study for a non-commercial purpose. I agree that a copy of the eThesis may be supplied to the British Library for inclusion on EThOS and WREO, if the thesis is not subject to an embargo, or if the embargo has been lifted or expired.		
5. I agree that the University of Sheffield's eThesis repository (currently WREO) will make my eThesis (where applicable) available over the internet via an entirely non-exclusive agreement and that, without changing content, WREO and/or the British Library may convert my eThesis to any medium or format for the purpose of future preservation and accessibility.		
6. I agree that the metadata relating to the eThesis (where applicable) will normally appear on both the University's eThesis server (WREO) and the British Library's EThOS service, even if the eThesis is subject to an embargo.		
Student's name (PLEASE PRINT): Lewis Hanney	Signature: 	Date 01/04/2020
SECTION 6: THESIS DEPOSIT AGREEMENT - SUPERVISOR		
I, the supervisor, agree to the named Thesis being made available in accordance with the conditions specified above.		
Supervisor's name (PLEASE PRINT): Professor Markus Reuber	Signature: 	Date: 01/04/2020

Declaration

I declare that this work has not been submitted for any other degree at the University of Sheffield or any other institution.

Word Count

Literature Review	**7,881**
Including references and tables	10,990
Research Report	**7,533**
Including references and tables	10,419
Total	**15, 414**
Including references and tables	21,409

Acknowledgements

Firstly I would like to thank both of my supervisors for their advice and feedback. I am thankful to FND Action and Epilepsy Action for their help and support with the study. I am very thankful to all of the participants who took part in the study, giving up their time. I learnt so much from working with you. I am grateful to Dr Jo Varela for her incredible support and Dr Claire Bone for her ceaseless good humour. I would also like to thank the ITU staff at QEQM, without them, none of this would have been possible. Finally, I would like to thank my family, in particular my girlfriend, my mother and grandmother for their support over a very difficult year and sticking with me through bouts of worry and ill humour.

Abstract

Stigma is when a certain characteristic (such as weight or race) causes an individual or group to be treated negatively based on a feature that differentiates them from other members of society. Stigma can be considered in different ways; Stigma can be 'enacted', when the individual is directly discriminated against, and 'perceived', when individual believes the negative actions of others to be linked to an undesirable characteristic of their self. Stigma can have serious consequences, such as reduced quality of life, lower rates of employment and less overall life satisfaction.

Epilepsy is a reasonably common neurological disorder where individuals experience seizures. Over 500,000 UK residents are estimated to be affected by Epilepsy. It can be caused by a number of different factors, such as brain lesions, but there may be no known cause for up to two-thirds of individuals with epilepsy. Historically, seizures have been associated with negative factors, such as demonic possession, and have experienced stigma. There is also evidence that stigmatising attitudes towards seizures continue to be prevalent.

Non-epileptic attack disorder (NEAD) is a condition whereby an individual experiences seizures that are outwardly similar to epileptic seizures. These seizures, however, are not associated with the neurological correlates of epilepsy. NEAD is a fairly common disorder with an estimated 15 000 UK residents receiving a diagnosis. The causes of NEAD are often considered psychological rather than physical and, as such, NEAD is often considered a mental health disorder.

The causes and manifestations of stigma in NEAD are unclear. This project aimed to extend the current understanding of stigma in NEAD by first: reviewing the literature on the experiences of stigma for NEAD and second: investigating the possible underlying factors that may cause perceived stigma in NEAD compared to epilepsy.

Part I describes the outcomes of a systematic narrative review that identified 22 papers investigating the experiences of stigma by individuals diagnosed with NEAD. The results indicate that individuals with NEAD experience stigma from healthcare professionals, the public, and family and friends. Some evidence suggests that individuals with NEAD experience higher rates of stigma than epilepsy.

Part II presents a cross-sectional study that investigate the relationship between perceived stigma and self-efficacy, depression, anxiety and seizure symptom severity in 2 groups (epilepsy and NEAD). The study found individuals with NEAD report more perceived stigma than those with epilepsy and higher, clinical, levels of depression. The severity of symptoms was not associated with perceived stigma for NEAD but it was with epilepsy. Depression was significant predictive factor for perceived stigma in both epilepsy and NEAD.

These studies develop the current understanding of stigma in NEAD and epilepsy. The findings suggest individuals with NEAD experience more stigma than those with Epilepsy and that depression predicts perceived stigma for other groups. The findings also suggest that stigma may be a useful target for future interventions for individuals with NEAD.

Contents

Part I: Literature Review

A systematic narrative review of the role of stigma in Non-epileptic Attack Disorder

Abstract

Objectives

To systematically explore the experiences of stigma from the perspective of individuals with Non-epileptic Attack Disorder (NEAD) and healthcare professionals involved in their care.

Method

A systematic narrative review of the literature was undertaken. Databases were searched by title, abstract and key terms. Searches were completed in MEDLINE, CINAHL, PsycINFO, SCOPUS, Web of Science and Google Scholar. The search was completed in July, 2019

Results

In total 22 papers were included in the final review. Experiences of stigma from the perspective of individuals with NEAD were found to be present in the literature. Where comparisons were made, individuals with NEAD reported higher levels of stigma than a comparable disorder (epilepsy). One paper suggested a possible treatment interventions for stigma in NEAD

There is emerging evidence for the considering importance of stigma in NEAD Further longitudinal research, however, would be beneficial.

Practitioner points

- Professionals should consider if their attitudes and terminology towards NEAD may result in individuals with NEAD feeling stigmatised.

- It could be beneficial to consider interventions targeting stigma in NEAD Focused Expressive Writing may be a useful intervention.

Limitations

- Many of the conclusions of the quantitative studies are largely based on correlational data from cross-sectional study. This limits our understanding of the causes of stigma in NEAD
- There is limited direct evidence of negative and stigmatising public attitudes towards NEAD, despite professionals and individuals with NEAD stating it as an important stigmatizing factor.

Introduction

Stigma is a process that can be defined as occurring when a specific characteristic of an individual is or appears to be devalued by others. It is often associated with thoughts of inadequacy, being rejected, and with feelings of shame and humiliation (Goffman, 1963; Link & Phelan, 2001). Existing research has examined stigma associated with a range of different conditions, including the quality of life in long term cancer patients (Johnson et al., 2019), the physical and psychological impact of limb amputation (Robert, 2019), and the visible appearance of skin conditions (Thompson et al., 2010), and help seeking behaviour of individuals with mental health disorders (Durna, Yorulmaz, & Aktac, 2019).

Goffman (1963) defined stigma as process by which a specific attribute of an individual is caused to be '*…reduced in our minds from a whole and usual person to a tainted, discounted one.*' (pg. 3). Goffman goes on to argue that a language of *'relationships not attributes'* (pg. 4) is needed, meaning that stigma is caused by how society relates to a specific attribute rather than a core or fundamental problem inherent in the attribute. Building on Goffman's original work, Link and Phelan (2001, pg. 363) argue that there are four core components that are required for stigmatisation to take place: Component 1: "The ability to distinguish and label differences"; Component 2: "Relating human differences with negative attributes" ; Component 3: "Separating "us" from "them""; Component 4: "status loss and discrimination".

Link and Phelan (2001) argue that the vast majority of human differences, whilst easily and readily observable, are ignored and therefore become socially unimportant. For example; differences between an individual's fingerprints or size of their ears are not major sources of stigma in the United Kingdom. However, the colour of an individual's skin or the colour of their hair, whilst arguably equally as arbitrary, can be sources of stigma resulting in

negative outcomes for the individual (Fernando, 2006; Kibria, 2008; Takeda, Helms, & Romanova, 2006). Historically, people who have experienced seizure have been viewed negatively (Wolf, 2010). Seizures have been linked to witchcraft and possession (Jilek-Aall, Jilek, Kaaya, Mkombachepa, & Hillary, 1997), and there is evidence that similar views continue to be prevalent (Millogo et al., 2019). Therefore, Link and Phelan argue, there is a 'social selection' (pg. 367) as to which human differences matter and therefore might be associated with stigmatisation and those that do not.

This construction of social differences relies on the cognitive oversimplification, a process by which individuals categorise and sort information into small, manageable chunks (Gigerenzer, 2008). Link and Phelan give the example of society dividing race into 'black' and 'white', whilst ignoring the nuances of ethnicity, culture and the different shades of skin colour.

Link and Phelan (2001) further note that labels which are deemed socially acceptable differ depending upon time and place. An example of this is given by Eknoyan (2006), who reports that being 'fat' was socially regarded as a positive, being a sign of success and wealth. Eknoyan charts the change in perceptions of being overweight from the nineteenth century change to finding the aesthetic of being overweight as unattractive to more recent times where being overweight can be regarded as both unattractive and as a sign of moral weakness.

As part of the second step of the process of stigmatisation, Link and Phelan argue that once an individual has been labelled, that person is then linked to a stereotype. In this context, a stereotype is a group of objectionable characteristics from which individuals want social distance. It has been further argued that stereotyping is a resource-preserving device (labelled 'Cognitive Efficacy' by Link & Phelan, 2001; see also Crocker & Lutsky, 1986).

This essentially means that stereotypes make it easier and quicker for human cognitive processes to compute and categorise large amounts of information, whilst maintaining other cognitive processes. Evidence for this comes from a study using a dual task paradigm, where participants had to form impressions of targets whilst monitoring prose. Participants showed improved prose-monitoring when stereotyped labels were used in the impression-forming task (Macrae, Milne, & Bodenhausen, 1994). Devine and Sharp (2009) further argue that stereotypes are not only resource-preserving, but also automatic and preconscious. Evidence for this has come from a number of studies. For example, Spencer et al. (1998) found that stereotype activation occurred following negative feedback after having glimpsed a face for a fraction of a second.

Link and Phelan discuss the third characteristic of stigmatisation as the use of social labels and stereotypes to split 'us' and 'them' (2001). The 'us' and 'them' dichotomy is a fundamental pillar of group psychology and whilst it has been argued that hostility towards outgroups is not always intentional (Brewer, 1999), it can often be the result (Weisel & Böhm, 2015). Whilst the make-up of the 'us' and the 'them' can change over time and in different circumstances (Hamilton, 2015), the core process of attributing negative and socially undesirable characteristics to 'them' remains (Hamilton, 2015). This attributing of negative and socially undesirable characteristics to out-groups can result in the members becoming dehumanised, which can be catalyst for perpetration of horrific acts (Haslam, 2006).

The fourth and final stage of Link and Phelan's (2001) model for stigmatisation is that the stigmatised person experiences loss of status and discrimination. Link and Phelan argue that this is a crucial, but also often neglected in the literature, stage of the stigmatisation process. When an individual is set-apart and associated with negative and unpleasant characteristics, a rationale develops for excluding them from meaningful societal inclusion.

For example, a study looking at the social media portrayals of individuals who experience seizures found that 41% of "tweets" were derogatory in nature (McNeil, Brna, & Gordon, 2012), essentially meaning that social media may be a difficult and threatening experience for NEAD. The consequences of social exclusion can be serious. Individuals who have been stigmatised and experienced status loss often perform poorly on matrices related to professional attainment, psychological functioning and life expectancy, amongst others (Livingston & Boyd, 2010; Mittal, Sullivan, Chekuri, Allee, & Corrigan, 2012).

Hatzenbuehler and Link (2014) have further developed this model and argue that stigma can at occur at different levels. For example; stigma can occur at intra-personal (stigma towards the self), the interpersonal (person-to-person stigma) and finally to the structural-level (also known as institutional stigma; organisational and governmental laws that result in stigmatisation). An example of structural stigma is the legal restricts on driving for people who experience seizures. A recent study from United States had indicated that seizures accounted for fatal car crashes less often (0.2%) than drunk driving (31%). Nevertheless, driving restrictions are common and a recent review focusing on driving and epilepsy found that driving restrictions are often listed as a major and important factor in the loss of independence.

Stigma can be further categorised into perceived and enacted stigma (also known as internal and external). Perceived stigma is when individuals attribute the negative action and behaviours of others to be related to a negative characteristic of their self. A powerful example of this is shown in the Kochman and Sikkema (2002) study that found HIV-positive sex-workers were reluctant to access appropriate medical interventions because of their feeling of being unworthy of such care. Additional, Research also suggests that stigma can negatively affect help-seeking behaviours. A systematic review by Clement et al. (2015) found that stigma regarding mental illness had a small- medium negative effect on help-

seeking behaviours. Concern surrounding making disclosures regarding the mental health problem were found to be the most frequently reported stigma barrier. They found that young men, individuals from ethnic minorities and professionals in the military and health-care settings were the most likely to be discouraged from seeking help because of the stigma. A further systematic review and meta-analysis by Schnyder, Panczak, Groth, & Schultze-Lutter (2017) had similar findings, although they suggest that it is the individual's personal attitudes rather than the broad public opinion, which should be targeted for intervention.

Delayed help seeking can have serious consequences for individuals with mental health difficulties (Dell'Osso & Altamura, 2010). For example, a study by Bukh, Bock, Vinberg, and Kessing, (2013) found that remission rates for individuals who had experienced a major depressive episode and had not received treatment for 6-months were significantly lower than for individuals who had been treated more quickly. A systematic review by Nordentoft et al. (2009) looking at the efficacy of early intervention for individuals with schizophrenia found that early intervention resulted in better treatment outcomes. Therefore, the result of delayed treatment because of stigma related to treatment seeking behaviour can have serious personal and social consequences.

In the context of the Link and Phelan's (2001) model, delayed treatment seeking demonstrate how they had internalised society's stigmatisation of them to the extent that it has become a core aspect of their personal narrative, preventing them from accessing lifesaving care (Yanos, Roe, West, Smith, & Lysaker, 2012; Yanos, Lucksted, Drapalski, Roe, & Lysaker, 2015).

External stigma is when individuals are directly discriminated against by others, be it at the inter-personal or societal level. For example; a review by Hughto, Reisner, and

Pachankis (2015) found that transgender people were less likely to be offered jobs and receive appropriate healthcare than non-transgender people.

The consequences of stigmatisation, as well as association loss of social standing and sense of power, has been further developed into a model for the development of mental health difficulties. The power threat-meaning framework (Johnstone & Boyle, 2018) posits that mental health difficulties develop as a consequence of fundamental inequalities in society, perpetuated by stigma, prejudice and discrimination. They argue that mental health difficulties are a response to experiences of powerlessness and associated threat. Examples of evidence for this comes to the finding that countries that have greater levels of social inequality and income disparity have higher prevalence of mental health difficulties (Pickett, James, & Wilkinson, 2006 Pickett Wilkinson, 2010).

Given the impact that experiences of stigma can have on physical and psychological health, it is unsurprising that researchers have also looked at the role stigma can play in the health of individual suffering with functional neurological disorders (FND) or Dissociative Neurological Symptom Disorder (DNSD) (e.g. Rommelfanger et al., 2017; Psychiatric Association, 2013). FND are conditions characterised by an involuntary inability to access motor, sensory, and/or cognitive functions. These symptoms are not related to the effect of disease or damage to the nervous system or caused by substance use (American Psychiatric Association, 2013; World Health Organization, 2018). FND are often regarded as a physiological manifestation of psychological difficulties (Ludiwick et al., 2018). Non-epileptic Attack Disorder (NEAD) is a specific type of FND in which the individual suffers from seizures, phenotypically similar to epileptic seizures, but without the neurological correlates of an epileptic seizure (Francis & Baker, 1999;Alsaadi & Marquez, 2005; Reuber, 2008). The causes of NEAD are unclear, although it has often been linked to maladaptive psychological coping (Brown & Reuber, 2016; Reuber & Brown, 2017). Some research has

highlighted that personality pathology is more frequent in individuals with NEAD (Reuber, Pukrop, Bauer, Derfuss, & Elger, 2004). In particular, they found that maladaptive personality traits similar to Borderline Personality Disorder were more common than in the general population or patients with epilepsy. Further research by Green, Norman and Reuber (2017) has indicated that individuals with NEAD have higher levels of anxiety and depression than individuals with epilepsy, as well as difficulties related to attachment styles. This, coupled with the lack of a known physical aetiology, results in individuals with NEAD meeting the classification for a disorder of mental health, with the exception being individuals who present with malingering seizures (American Psychiatric Association, 2013). Many individuals with NEAD described the confusion between their diagnosis and epilepsy, even from health care professionals (Auxemery, Hubsch, & Fidelle, 2011) which can result in the construction of social differences can also be related to NEAD.

NEAD is often diagnosed following a misdiagnosis of epilepsy and a subsequent lack of response to epilepsy specific treatment. A diagnosis of NEAD requires specialist testing (which involved video-electrographic, VEEG, recording of typical seizures) because it can be difficult to outwardly differentiate between epileptic and non-epileptic seizures. It is possible that an individual may be misdiagnosed with NEAD and actually be suffering with epilepsy, but there is little evidence for this in the available literature.

Most non-epileptic seizures are perceived by patients as outside of their own control and, similarly to epilepsy, impair the individual's normal sensory and cognitive functioning. Given the outwardly similar appearance of NEAD and epileptic attacks, individuals with NEAD are often initially misdiagnosed with epilepsy, and only receive the accurate diagnosis of NEAD after a delay of several years. Furthermore, it is recognised that epilepsy is a risk factor for the development of NEAD and some individuals will receive a mixed diagnosis of epilepsy and NEAD, although this is relatively uncommon and the vast majority of patients

either have epilepsy or NEAD (Wissel et al., 2016). Specialised testing, including video-electroencephalographic recording of typical seizures and clinical assessment by an expert may be required to differentiate between non-epileptic and epileptic seizures.

NEAD is three to four times more common in women than men, with the onset of symptom frequently occurring in the teens or early 20s, although seizures can start at any age (Mellers, 2005). There are an estimated 15,000 people with a diagnosis of NEAD in the UK, with an estimated prevalence rate of 50 per 100,000 (Kanemoto, et al. 2017).

The relationship between stigma and mental health disorders is well established (Clement et al., 2015; Schulze, 2007; Sharac, Mccrone, Clement, & Thornicroft, 2010). Research suggests that individuals with a diagnosis of a mental health disorder report experiencing more stigma than those with a physical health conditions, for example Glozier, Hough, Henderson, and Holland-Elliott (2006) found that psychiatric nurses were less sympathetic to colleagues returning to work after sick-leave related to mental health than they were to colleagues returning to work after physical illness (diabetes). The impact of stigma for individuals with mental health difficulties can also have serious consequences. A systematic review by Sharac, McCrone, Clement, and Thornicrof (2010) found that mental health related stigma results in fewer employment opportunities, lower income and less resource allocation to mental health related care. Interestingly, the authors propose that not only would reducing mental health stigma be beneficial for the individuals, but also that society, as a whole, would gain a positive economic benefit. Another example of the stigma that individual's with mental health difficulties experience is 'Diagnostic-overshadowing'. This is the tendency of physical symptoms to be misattributed to mental health difficulties.

As with many mental health disorders, individuals with NEAD have reported experiencing stigma related to their disorder, at an even higher rate than individuals with epilepsy (Rawlings, G. Brown, & Reuber, 2017). There have been a number of studies examining stigma in NEAD populations, but as of yet no comprehensive review of the available literature has been conducted. Given the detrimental impact of stigma on quality of life (MacLeod, & Austin, 2003) and increased psychological distress (Earnshaw, & Quinn, 2012), the present review aims to provide a better understanding as to how individuals with NEAD experience stigma, especially at diagnosis. Therefore, the present systemic narrative review aims to systematically identify studies that have investigated the role played by stigma in the lives of individuals with NEAD.

Method

Search strategy

Database search: Databases were searched by title, abstract and key term. Searches were completed in MEDLINE, CINAHL, PsycINFO, SCOPUS, Web of Science and Google Scholar. Reference sections of the final papers were examined, and a forward citation search was completed. The search was conducted in July, 2019.

Inclusion criteria:

If studies were in English and in a peer-reviewed journal, then their titles and abstracts were scanned and included if appropriate. Papers were included is they related to individuals with NEAD or healthcare professionals discussing stigma, were focusing on measuring stigma for individuals with NEAD or were regarding an intervention related to stigma for individuals with NEAD or the public.

Exclusion criteria: Studies were excluded if they made no mention of stigma experienced by individuals with NEAD or the attitude of healthcare professionals towards NEAD in relation to stigma were not related to the measurement of stigma in individuals with NEAD or interventions related stigma in individuals with NEAD.

Search terms: Search terms relating to stigma and NEAD were used including: stigma, stigmatisation, NEAD, PNES and functional seizure disorder (see appendix A for full list of search terms).

Data coding and extraction. A data extraction and coding scheme (see Appendix B) was developed to extract important information from the final sample of studies. These included the author, publication date, country of origin, sample demographics, methodology (qualitative, quantities, mixed), measures and outcomes (if appropriate), statistical procedures (if appropriate), findings and conclusions. The resulting information was extracted and entered into a database. The primary researcher then interpreted and synthesised the data to address the research questions.

Quality appraisal. Quality appraisal checklists from the Joanna Briggs Institute (JBI, 2019) were used. Different JBI tools were used depending upon the study type. For qualitative research, the "Checklist for Qualitative Research was used" (appendix C). For cross sectional studies, the "Checklist for Analytical Cross Sectional Studies" was used (appendix D). For randomised controlled trial (RCT) the "Checklist for Randomized Controlled Trials" (See appendix E). The JBI checklists cover between 8-13 questions that help the reader to consider different aspects of the quality of the paper, including potential bias and appropriateness of research methodology. The overall quality of each study was calculated by combining the scores form each item on the checklist and converting them into a percentage to aid comparison between study types. Higher scores indicted the greater

quality of a study (see appendix F for full rating breakdown). A second rater (trainee clinical psychologist) was used to check the reliability of the ratings. Cohens Kappa was used to rate interrater reliability, which resulted in "moderate agreement" (*k*= .49 (p<.001), 95% CI = .41 - .57). Any disagreements were discussed jointly and a final rating agreed upon. JBI does not indicate a minimum score by which papers should be excluded, therefore no papers were excluded based on their checklist sores. However, the checklist scores were combined to interpret the finding of the study.

Results

All of the databases were searched systematically using the identified search terms, resulting in the retrieval of 2310 papers. Any duplicates were excluded. The remaining papers were screened by title for relevance, applying the inclusion/exclusion criteria. Forward and backwards citation search of the remaining articles were conducted. In total, 26 full-text articles were considered. Four were excluded for reasons provided in figure 2. This resulted in the inclusion of 22 for the final synthesis. All of the included were papers were screened using the JBI checklists with the scores also found in figure 2.

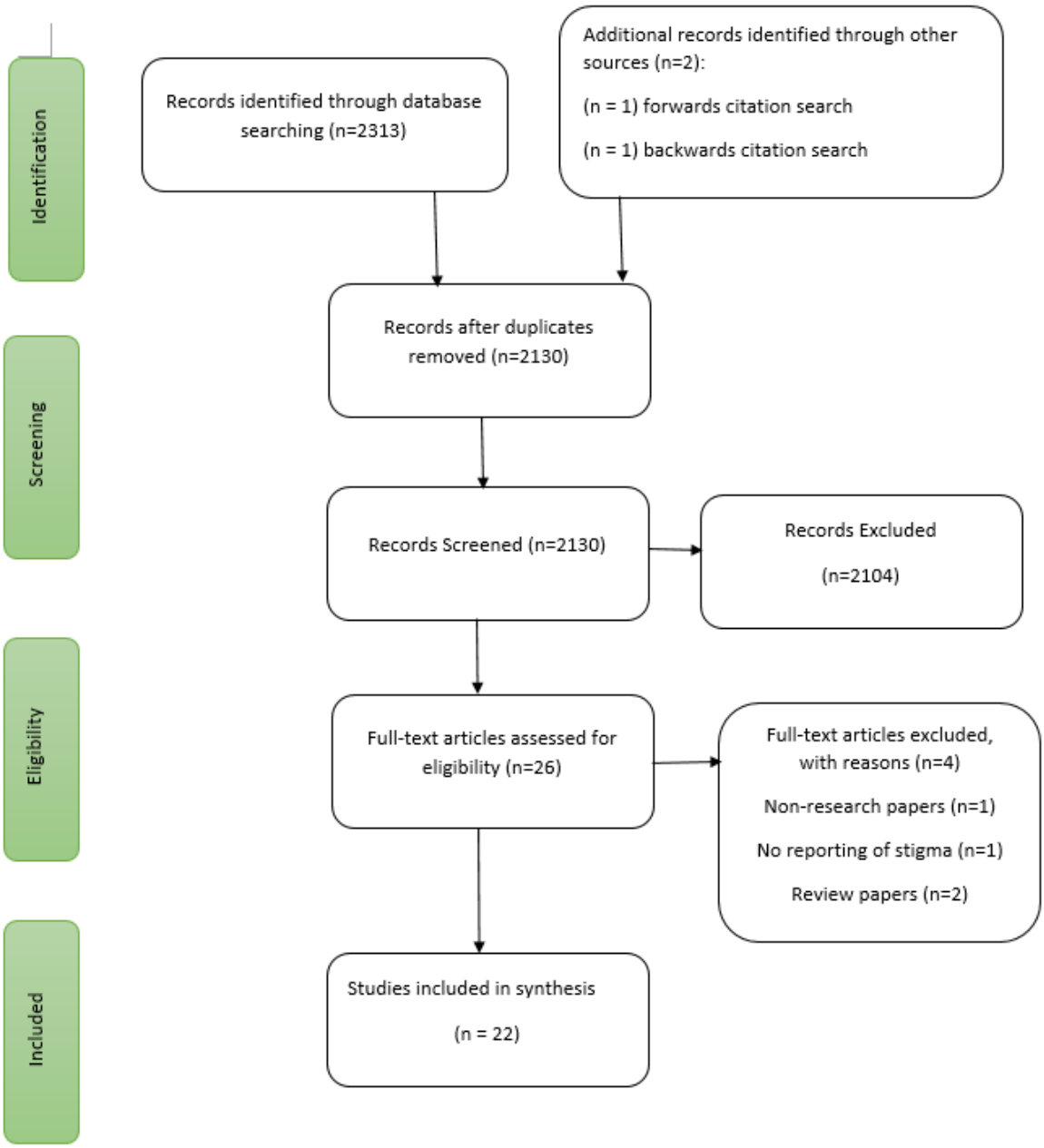


Figure 2. PRISMA flow diagram

	Primary author	Date	Country	Sample size/Characteristics	Design	Methodology	JBI Score
13	Carter, Denton,Ladino, Hassan, Sawchuk, Snyder... & Group.	2018	Canada	N = 62 Healthcare professionals	Structured questionnaire	Quantitative	87.5%
14	Carton, Thompson & Duncan	2003	UK	N = 84 individuals with NEAD over the age of 18	Structured questionnaire	Quantitative	50%
1	du Toit, & Pretorius,	2017	Nambia	N=15 Healthcare Professional	Semi-structured interviews using thematic analysis	Qualitative	80%
2	Fairclough, Fox, Mercer, Reuber, & Brown	2013	UK	N=12 individuals with NEAD over the age of 17	Semi-structured interviews	Qualitative	80%
15	Hingray, El Hage, Duncan, Gigineishvili, Kanemoto, LaFrance Jr, & Wiseman	2018	Global	N=1098 Healthcare professionals	Structured questionnaire	Quantitative	87.5%
3	Karterud, Knizek, & Nakken	2015	Norway	N =11 Individuals aged 14-23 with a diagnosis of NEAD	Semi-structured interviews	Qualitative	80%
4	Karterud, Haavet, & Risør.	2016	Norway	N = 14 – 24year with a diagnosis of NEAD	Semi-structured interviews analysed using thematic analysis.	Qualitative	90%
5	McWilliams, Reilly, McFarlane, Booker, & Heyman	2016	UK	N = 10 young people 0-19 and 29 family members	Semi-structured interviews using thematic analysis for the analysis	Qualitative	80%

	McMillan, Pugh,	2014	USA	N= 74 Healthcare	Semi-structured	Qualitative	80%
			US and Canada	NEAD over the age of 18			
11	Robson & Lian	2017	UK, Ireland, US and Canada. Australia, New Zealand and Norway	N=135 individuals with NEAD over the age of 18	Structured questionnaire using thematic analysis to analyses free text	Qualitative	80%
18	Tong, An, Reuber, Zhang & Zhou	2018	China	N = 102 Healthcare professionals	Structured questionnaire	Quantitative	87.5%
19	Vaidya-Mathur, Myers, Laban-Grant, Lancman, Lancman, & Jones	2016	USA	N= 141 individuals with NEAD over the age of 18	Structured questionnaires	Quantitative	75%
20	Worsely, Whitehead, Kandler, & Reuber	2011	UK	N=61 healthcare professionals not including Doctors	Structured questionnaires (Illness Perception Questionnaire—Revised (IPQ-R)and the single-item Symptom Attribution Question (SAQ))	Quantitative	75%
12	Wyatt, Laraway & Weatherhead	2014	UK	N=6 individuals with NEAD over the age of 18	Semi-Structured Interviews using thematic analysis	Qualitative	80%
21	Yogarajah, Child, Agrawal, Cope, Edwards, & Mula	2019	UK	N=120 London based GPs	Structured questionnaire	Quantitative	75%

Study Characteristics

Detailed study characteristics can be found in table 1. Twenty-six full-text papers were considered for inclusion and four of these were excluded for reasons provided in figure 1. This resulted in 22 papers being included in the review. Of the 22 articles 9 reported cross-sectional studies, one reported a randomised controlled trial and 12 reported qualitative studies. One sample was analysed in two separate studies (Rawlings, Brown, Stone, & Reuber, 2018; Rawlings, Brown, & Reuber, 2019), however both were included because they looked at the participant data using different qualitative methodology and covering different topics.

Quality appraisal

All papers were checked for quality using the appropriate JBI quality checklists (see appendices C, D, E). All studies were included regardless of quality appraisal and the appraisal was used for informational purposes only and to facilitate commentary on the overall strengths and weaknesses of the research area. The mean quality percentage for all studies was 80%, with a range of 50-90%. For quantitative studies, the mean was 79%, with a range 50-87.5%. For qualitative studies, the mean was 81%, with a range of 70-90%.

Stigma and NEAD

All of the studies included discussed the role of stigma in individuals with NEAD. Upon examination, the articles can be divided into the following themes:

Adjusting to diagnosis: Five studies examined how individuals adjusted to a diagnosis of NEAD. Being diagnosed with NEAD, especially for those who had previously been diagnosed with epilepsy, was a common source of stigma (Wyatt, Laraway & Weatherhead, 2014; Pretorius & Sparrow, 2015; Pretorius; Karterud, Knizek, & Nakken, 2015). Wyatt, Laraway, and Weatherhead (2014) interviewed six individuals with NEAD,

using Thematic Analysis (TA) to highlighted concerns that many people felt that psychological treatment would not be any more effective than anti-epileptic drugs (AED), and was another hurdle before reaching a final diagnosis or help. Another theme found in several studies to be associated with the diagnosis process was the sudden change from a "physical" to a "psychological" disorder, and many studies indicate stigma was associated with the shift to use a "mental health" label (Wyatt, Laraway, & Weatherhead, 2014; Pretorius & Sparrow, 2015). This is exemplified by "Lisa" in Wyatt, Laraway, and Weatherhead's (2014) study, where one of the six interviewed participants believed that a diagnosis of NEAD would result in her being detained in a "psychiatric hospital" (pg. 405). Pretorius and Sparrow (2015), who investigated adjustments to diagnosis' of NEAD in a South African population, found that many participants found a psychological cause to be stigmatising and that there was also an underlying belief that psychological problems were less "real" (pg. 36) than physical problems. Similarly, questionnaire based studies by Tong, An, Reuber, Zhang, and Zhou (2018), found that many HCPs in China reported that many individuals diagnosed with NEAD declined the support of mental health services because of the stigma associated with having mental health difficulties. McMillian, et al. (2014) conducted a large qualitative study with 74 HCPs who worked with veterans in the United States and found that they too found that many individuals with NEAD declined mental health services because of the stigma of having a mental illness. Similarly, Du Toit and Pretorius (2018) found that many individuals in Namibia struggled with being diagnosed with NEAD, predominantly because of the cultural stigma attached to mental health difficulties. This cultural stigma often stopped or delayed, individuals seeking psychological treatment.

Pretorius (2016), also studying the experience of South Africans who had been diagnosed with NEAD, found that the attitude of some healthcare professionals (HCP) at diagnosis was a cause of the feeling of stigma. This included being told that the individual

was '*faking it for attention'* (pg.3) and that the many HCPs were less supportive and empathetic when the cause of the seizures was attributed to "*mental health*" rather than "*a medical issue*" (pg. 3).

Attitudes of HCP: Four papers looked at the attitudes of HCPs towards NEAD. Yogarajah, Child, Agrawal, Cope, Edwards, and Mula (2019) sent questionnaires to 974 London GPs regarding NEAD, of which 120 replied. They found that almost 75% continued to use the term "*pseudo-seizures*", despite the pejorative connotations this term may have and the possible stigma it may cause (Barron, & Rotge, 2019). Indeed, a small number (n=8) of GPs continued to use the term "*hysterical seizures*", despite this being regarded as "*highly offensive*" by individuals with NEAD (Dunne, Carolan, Swords, & Fortune, 2019). Additionally, this study highlighted that over 50% believed that the seizures were voluntary, again reinforcing the stigma that because individuals with NEAD lack the physical correlates of epileptic seizures, they have more volition over their seizures. These stigmatising attitudes may be a product of inexperience of supporting individuals with NEAD, with 89% of the General Practitioner (GP) who replied saying that they have seen less than 10 individuals with NEAD and approximately 50% who felt uncomfortable managing individuals with NEAD being "younger" GPs, who may have less experience than the "older" GPs. A cross-sectional survey by Carton, Thompson & Duncan (2003), found that some GPs did not believe the diagnosis, despite being diagnosed by a specialist centre. They contacted the GPs of 84 newly diagnosed NEAD patients and, at follow-up, found that 10 of the GPs did not agree with the diagnosis and continued to prescribe potent Anti Epileptic Medications. The reason for this disagreement is not clear, but may be linked to the stigmatising attitude toward NEAD not being a valid disorder. It should be noted, however, that this study had the lowest percentage of all studies included in the review and suffered from a lack of clarity regarding methodology.

A qualitative study using Grounded Theory by McMillian et al. (2014) also found that the attitudes of clinicians could be stigmatising. This study, which took place in the United States in Veterans Associations clinics, interviewed 79 HCPs (such as neurologists and nurse practitioners). Many of the HCPs interviewed displayed stigmatising attitudes towards individuals with NEAD. A major theme was that individuals with NEAD were malingering or trying to maximise their opportunity for disability benefits. A high-quality qualitative study by Du Toit and Pretorius (2018), looked at the attitudes of HCPs in Namibia, found that many HCPs reported stigmatising attitude of their peers. They reported that many HCPs did not regard NEAD as a valid disorder, and one participant highlighted an example of a patient being refused hospitalisation by a medical aid because the individual did not regard NEAD as a disorder that would ever require hospitalisation. Stigmatising attitudes were also displayed by some respondents themselves. For example, they highlight that one participant stated that "*I don't think that they* [the public*] know that there can be a difference between real seizures and pseudo-seizures*" (pg. 50). This comment again uses the stigmatising and invalidating terminology of 'pseudo-seizures' but also reinforces the unhelpful dichotomy of real seizures vs. not real seizures, as highlighted by David (2012.)

Worsely, Whitehead, Kandler, and Reuber (2011) also investigated the attitudes of HCPs (although not including medical doctors) towards individuals with NEAD. This high-quality cross-sectional quantitative study used the adapted Illness Perception Questionnaire-Revised and the Symptom Attribution Questionnaire for epilepsy and PNES. They found that most HCPs believed that individuals with NEAD had control over their seizures and that NEAD was less chronic than epilepsy. Again, these views minimise the difficulties faced by individuals with NEAD and reinforces the stigma associated with this disorder. Tong, An, Reuber, Zhang, and Zhou (2018) also quantitatively reviewed the attitudes of HCPs, although focusing on the views of HCPs in urban China. Although this research did not specifically

address stigmatising attitudes of HCPs, it did find that neurologists would invite less than half (41%) of individuals with NEAD back for a follow-up appointment following diagnosis, and that many of those invited back may have co-morbid epilepsy. The reason for the lack of follow-ups is unclear, but may be linked to NEAD not being regarded as a "medical" condition in urban China and therefore not suitable for further medical treatment. The lack of professional experience of managing NEAD by HCPs, and the resulting stigma, was highlighted by Higray et al. (2018). In this global quantitative study, they found that many HCPs believed that a lack of education related to NEAD resulted in stigmatising attitudes and interactions with patients.

Experiences of Stigma. A high quality cross-sectional study by Robson, Myers, Pretorius, Lian, and Reuber (2018) found that many individuals with NEAD experienced stigma. They used the "Epilepsy Stigma Scale" to measure stigma and found that individuals with NEAD reported experiencing higher than average levels of stigma. They also found a negative correlation with health-related quality of life (as measured by the "Quality of Life in Epilepsy"), indicating a link between stigma and quality of life. A study by Rawlings, Brown, and Reuber (2017) also found that individuals with NEAD are 42% more likely to report stigma than those with epilepsy. They also found that whilst stigma was correlated with symptom severity for epilepsy, it was not with NEAD. This suggests that individuals' perception of their disorder, influenced enacted and structural stigma, may be more important in the perpetuation of stigma than the disorder's physical manifestation.

This sense of stigma regarding their disorder is also highlighted by Vaidya-Mathur (2018), in a quantitative study based in the United States, looking at the socialisation characteristics of individuals with NEAD. This study found that 12% of respondents stated that social stigma related to their disorder stopped them from socialising. Unfortunately, this study did not have a comparison group, so it is difficult to place with a percentage in context

compared to other disorders. However, it may be telling that 41% of respondents reported being single compared to a national average of 26.9% and that 30.5% of individuals with NEAD reported that they were married compared to a national average of 56.4%. A narrative analysis by Rawlings, Brown, and Reuber (2018) also found that social stigma was a component in one of the important narratives that emerged from writing about living with NEAD. Social stigma was an important aspect of "tackling adversity" and participants discussed difficult social interactions, such as others saying that the individual with NEAD was faking a disability. Whilst the participant suggested that this specific interaction was said in jest, they also expressed hurt and disappointment at this attitude.

The theme of social stigma also emerged from a qualitative study by McWilliams, Reilly, McFarlane, Booker, and Heyman (2017). This study reported on the stigmatisation faced by young people with NEAD and their families. Several young people and their families reported that they had faced stigma from schools, including the school asking for the young person to not attend because of NEAD. This resulted in some young people being home schooled and missing out on important social and educational opportunities. The impact of stigmatisation, social isolation and the subsequent negative impact on emotional development has been documented in young people with epilepsy (Hightower, Carmon, & Minick, 2002) and this study by McWilliams, Reilly, McFarlane, Booker, and Heyman, suggests that the consequences for young people with NEAD may be the same. Karterud, Haavet, and Risø (2016) also highlight the role of social stigma in social isolation for young people. This qualitative study which took place in Norway, particularly emphasised that the belief that non-epileptic seizures were 'fake' as a significant source of stigma. They also discuss how NEAD could inhibit a young person's ability to access employment, exacerbating the "us and them" dichotomy and increasing social isolation.

Experiences of stigma from HCPs. Robson and Lian (2017) conducted a large cross-sectional qualitative study looking at how individuals with NEAD in the UK, US, Ireland and Canada had experienced interactions with HCPs. Many respondents reported that HCPs often defined NEAD as not being epilepsy. This parallels with Link and Phelan's (2001) first step of the stigmatisation process: "The ability to distinguish and label differences". Individuals with NEAD felt that they had been clearly labelled as different and that the label had been defined in the context of a lack of epilepsy. Participants further felt that NEAD had been linked with negative connotations (such as malingering, lazy and faking for the purpose of disability fraud), which further parallels component 2 of Link and Phelan's model ("Relating Human differences with negative attributes"). As highlighted by Robson & Lian, this can result in an "empathy gap" emerging for HCPs towards NEAD. This may result in the creation of a dichotomy between patients deserving and not deserving of empathy (Component 3 "Separating "us" from "them""). This can result in a myriad of difficulties (Component 4: "Status loss and discrimination"), ranging from an unwillingness to seek appropriate treatment (as highlighted by the papers mentioned above) to the high rates of unemployment as evidenced by 65% of respondents for this study being out of work**.**

A qualitative study, by Rawlings, Brown, Stone, and Reuber (2018a) using written accounts of living with NEAD reports experiencing stigma from HCPs by individuals with NEAD. This study compared written accounts of individuals with Epilepsy and with NEAD. They found that almost all participants with epilepsy reported positive interactions with HCPs, but those with NEAD reported a significant number of negative interactions, including HCPs not believing their symptoms and/or believing that those with NEAD had control over their seizures. This is an example of Component 3 ("Separating "us" from "them"") of Link and Phelan's (2001) model of the process of stigmatisation. It seems that many individuals with NEAD feel that they have been separated from being a patient deserving of care to

patient who is less deserving of care. Some participants with NEAD reported that the stigma of their disorder and experience of negative interaction with HCPs had resulted in them avoiding health care services. Similarly, McWilliams, et al. (2017) found that many young people and their families had negative experiences from HCPs and schools. Similar themes emerged from a study by Fairclough, Fox, Mercer, Reuber, and Brown, (2013), looking at the perceived treatment needs of individuals with NEAD. They found that the stigmatising attitudes of some HCPs (such as accusations of faking etc.) resulted in confusion and ambivalence regarding treatment, particularly towards psychotherapeutic interventions.

Public attitudes towards NEAD. Three studies highlight the poor public understanding and potentially stigmatising attitudes towards NEAD. Du Toit and Pretorius (2018) highlight that many HCPs regard the public understanding of NEAD in Namibia as being very poor. Some argue that the lack of public knowledge regarding NEAD can act as a barrier to treatment seeking and result in friends and family not being as supportive as they would be with other disorders. A large, high quality quantitative study by Carter et al. (2018) also found that many HCPs felt that a lack of public awareness of NEAD resulted in stigmatisation from both the general public and HCPs. In particular, many HCPs agreed that a lack of widespread understanding of the psychological mechanisms of NEAD delayed or stopped the use of psychotherapeutic treatment. Higray et al. (2018), in their large global study, found that many HCPs believed that a major barrier for diagnosis, treatment and causes of NEAD, was a lack of popular awareness of stigma.

Treatments for stigma in the NEAD population. There is only one study looking at the role of stigma in possible treatment options. A RCT by Rawlings, Brown, Stone, and Reuber (18b), investigated the role of focused expressive writing (FEW), where the individual writes about distressing events for approximately 15-20 minutes. A study by Lepore, and Greenberg (2002), suggests that FEW may be particularly useful treatment for

individuals who experience shame and stigma. The study uses the Quality of Life in Newly Diagnosed Epilepsy (NEWQOL-6D), a measure of health-related quality of life, which includes questions related to stigma. The control group had to write about their actions and behaviours, whilst the treatment group had to focus on four 'therapeutic' topics. The results indicated that individuals in the treatment group showed a significant positive increase in their NEWQOL-6D scores than the control group. Whilst this treatment may not be specific to stigma, it may be a possible mechanism of change for potentially improving quality of life for individuals with NEAD.

Discussion

This review has aimed to investigate how people with NEAD experience stigma, if the attitudes of HCPs towards NEAD contributes to stigma, and if stigma has been targeted for treatment in this population. Overall, a total of 22 studies were included for this review. JBI critical appraisal tools were used to check the quality of the papers. JBI scores ranged from 50% (which equates to acceptable quality) to 90% (very good quality). Ten of the studies used quantitative methodology and 12 qualitative methodology. Only two of the quantitative studies directly investigated stigma in NEAD, suggesting that this might be an under-researched area compared to epilepsy (Jacoby, 2008; MacLeod, & Austin, 2003). A wide range of different measures were also used to assess stigma in HCPs and those with NEAD, but none were specially validated in a NEAD population, with many using measures designed/validated in epilepsy as proxies.

Evidence suggested that individuals with NEAD report more stigma than individuals with epilepsy (Rawlings, Brown, & Reuber, 2017). This support research by Looper and Kirmayer, (2004), which found that individuals with functional disorders reported higher levels of stigma than individuals with comparable medical disorders. There is further

evidence from this review that individuals with NEAD report high levels of stigma and that stigma is negatively correlated with quality of life. Research suggests that lower quality of life is correlated with developing mental health difficulties (Alonso, et al., 2004). This suggests that the high levels of stigma experienced by NEAD, negatively impacts on quality of life, which may result in the development of further mental health problems.

This could be particularly concerning, given that 6 papers suggested that the attitudes of HCPs, particularly at diagnosis, resulted in individuals with NEAD reporting that they would be less likely to seek support from HCPs, or attend appointments, in the future. In particular, the papers highlighted that the change from a 'physical' to a 'mental' illness was particularly difficult for individuals with NEAD. This is supported by evidence that stigma towards mental health problems is a significant factor in delayed treatment seeking behaviours (Schomerus, & Angermeyer, 2008; Schnyder, Panczak, Groth, & Schultze-Lutter, 2017). The consequences of delayed treatment seeking can be serious, including an increase in the severity and duration of experiencing distress, and lower responsiveness to treatment (Bukh, Bock, Vinberg, and Kessing, 2013; Clement et al., 2015). Given the treatment gaps highlighted by Kanemoto et al. (2017), this delay in NEAD treatment could be particularly serious.

Only one study looked at the role of stigma in treatment options for NEAD. Although not specifically focused on stigma, the study did find an improvement on NEWQOL-6D (which contains items focused on stigma) following 15-20 minutes of directed focused expressive writing (FEW). Smyth and Helm (2003) found that FEW was an effective and inexpensive, self-help treatment for individuals who had experienced trauma. Given the possible aetiological links between NEAD and trauma (Reuber, 2008), FEW may prove to be an effective and inexpensive treatment options for individuals with NEAD to reduce stigma and improve quality of life.

Six articles also suggest that individuals with NEAD experience stigma from professionals after diagnosis. One study compared written accounts of living with NEAD to epilepsy and found that individuals with NEAD reported more negative and stigmatising interactions with HCPs then those with epilepsy. Research by Hederson et al. (2014) that many health professionals display stigmatising attitudes towards individuals with mental health problems. This includes questioning the legitimacy of the lived experience of the individuals with mental health problems (Corrigan, & Wassel, 2008). The reviewed literature suggests that this is a similar experience of individuals with NEAD, with many HCPs questioning the legitimacy of the diagnosis or regarding the seizure as 'fake'.

The description of poor care reported by individuals with NEAD is supported by research. There is evidence that individuals with mental health difficulties often received poor health care compared to individuals with physical health problems (Jones, Howard, & Thornicroft, 2008). This has often been linked to 'diagnostic overshadowing'; when HCPs attribute health related problems to a single diagnosis, such as mental health or learning disabilities. 'Diagnostic overshadowing' may be a particular problems in functional disorders, given the already unclear aetiology, and the associated stigma (Shefer et al., 2015).

Four papers also looked at the attitudes of HCPs themselves and found that many HCPs expressed attitudes that could be regarded as of a stigmatising nature. This included the continued use of outdated terminology (such as "pseudo-seizures" or "hysterical seizures to describe NEAD). Another finding was that many HCPs regarded NEAD seizures as often being voluntary, and as a mental and not physical disorder. It has been argued that the dichotomy between true illness and malingering is more relevant than the dichotomy of mental or physical illness. David (2012) argues that voluntary vs. involuntary is a more helpful way to regard NEAD seizures. An involuntary seizure is when the individual has no control over when they seize or how long they seize for. Epileptic and the majority of NEAD

seizures would fall into this category. A voluntary seizure would be when an individual has control of when they present with seizure like symptoms and would often be in the context of medical malingering. David argues that the aetiology of the seizures (be it physical or psychological) is important in the context of treatment but otherwise is irrelevant if the seizures are involuntary. By also regarding, the seizures as involuntary and not simply the product of mental health difficulties may help to reduce the stigma some HCPs have towards mental health difficulties (Henderson, et al., 2014).

Many individuals with NEAD, and HCP, highlighted the lack of knowledge in HCPs and also the general public. Work by Hederson et al. (2014) suggestions that improving educational resources for HCPs on mental health difficulties can help to reduce stigmatising attitudes. Three studies highlighted that public attitudes towards NEAD was also commonly regarded by HCPs as a source of stigma and a potential barrier to treatment. The link between negative public attitudes towards a disorder and delayed treatment seeking and poor outcomes is also supported by the research for mental health difficulties (Mojtabai, 2011).

Limitations

The outcomes of this review should be interpreted in the context of a number of limitations. Firstly, all of the papers included were published in English. They were also all drawn from peer-reviewed journals. Therefore, the included articles may not be a comprehensive reflection of the available literature of stigma and NEAD. It also opens the review to the possibility of publication bias, given that studies showing a positive link between NEAD and epilepsy were more likely to be published. It should be noted that the grey literature was searched, but no un-published work was found. It may, therefore, be that contrary findings might exist.

Secondly, many of the quantitative studies are of a correlational nature. Therefore, it is difficult to establish causality. No longitudinal studies looking at NEAD and stigma pre- and post-diagnosis of NEAD. Therefore, the high levels of stigma found in NEAD may have been present before the diagnosis and could have been linked to the aetiology of the disorder. Additionally, there was a lack of research exploring the link between stigma and other factors such as depression and anxiety. There is also little research on whether reducing stigma is a useful mechanism-of-changed for treatment Future longitudinal research focusing on stigma is warranted, particularly charting stigma pre-and post-diagnosis and pre- and post-treatment.

Whilst many of the studies were of a high quality, the investigators positionality and reflexivity were rarely stated in the qualitative. It is therefore difficult to consider any potential influences the investigators may have had on the participants, data and interpretation. Additionally, how this bias was accounted for in the results in rarely discussed.

Another limitation is that whilst many HCPs and individuals with NEAD stated that they perceived public attitudes towards NEAD to be a source of stigma, there was no direct evidence of this. A study investigating the awareness and attitudes towards NEAD in the context of stigma would be useful to establish public attitudes towards NEAD. This would help establish the level of stigma towards NEAD, direct and target any intervention to reduce negative public attitudes towards NEAD and allow a greater understanding of the mechanisms of stigma in NEAD.

Finally, given the variation in methodology and measurement of stigma, a meta-analysis was not completed. Therefore, researcher bias could influence the finding from this review. Although attempts to minimise bias through the use of objective coding and checklists, the outcomes of the review could have been influenced by the primary reviewers

own positionality. A future review could include more researchers in the interpretation and synthesis, which may help to reduce this bias.

Clinical implications

Despite the limitations highlighted above, a number of clinical implications can be drawn from this review. Many individuals with NEAD reported experiencing stigmatising attitude and terminology from HCP. Therefore, it is important for HCPs to be aware of how they are being perceived by individuals with NEAD and to be aware of current literature related to non-stigmatising terminology. It may be helpful for HCPs to consider the voluntary vs. involuntary diagnosis, rather than regarding NEAD in the context of a physical/mental health dichotomy. It may also be beneficial for HCPs to improve their understanding of NEAD given its relatively high prevalence, (Reuber, 2008), and to promote education of NEAD among peers and the public. This may help to reduce stigma towards NEAD and reduce barriers to treatment seeking.

In terms of direct treatment options, it may be beneficial for psychotherapeutic interventions to consider the role of stigma in NEAD, especially in the context of quality of life. Research stated in the qualitative studies suggests that psychotherapy can be effective in reducing self-stigma (Wykes, & Hayward, 2006; Macinnes, & Lewis, 2008). Therefore, psychotherapy may be effective in reducing self-stigma in NEAD and improving quality of life. The use of FEW for NEAD may also be effective and should be considered as a possible treatment option, especially given that it would be an inexpensive intervention.

Conclusion

In conclusion, the current review suggests that individuals with NEAD experience stigma related to their disorder. They report that they experience stigma from both HCPs, friends and other organisations such as schools. Research suggests that FEW may be a useful

intervention for reducing stigma in NEAD. These findings should be considered in the context of possible researcher and publication bias. Therefore, further investigation of this topic is warranted. In particular, longitude studies investigating NEAD pre-/post-diagnosis and pre-/post-treatment would help to better evaluate the relationship between stigma and NEAD and establish additional mechanisms of reducing stigma.

References

Alonso, J., Angermeyer, M. C., Bernert, S., Bruffaerts, R., Brugha, T. S., ... & Gasquet, I. (2004). Disability and quality of life impact of mental disorders in Europe: results from the European Study of the Epidemiology of Mental Disorders (ESEMeD) project. Acta Psychiatrica Scandinavica, 109, 38-46. https://doi.org/10.1111/j.1600-0047.2004.00329.x

Alsaadi, T. M., & Marquez, A. V. (2005). Psychogenic nonepileptic seizures. *Am Fam Physician, 72*, 849-856. doi: 10.1007/s10072-017-2887-8.

Auxemery, Y., Hubsch, C., & Fidelle, G. (2011). Psychogenic non epileptic seizures: a review. *L'Encephale, 37,* 153-158. doi: 10.1007/s10072-017-2887-8.

Barron, E., & Rotge, J. Y. (2019). Talking about “psychogenic nonepileptic seizure” is wrong and stigmatizing. *Seizure-European Journal of Epilepsy*, *71*, 6-7. https://doi.org/10.1016/j.seizure.2019.05.021

Bastian, B., & Haslam, N. (2006). Psychological essentialism and stereotype endorsement. *Journal of Experimental Social Psychology, 42*, 228-235. doi: 10.1016/j.jesp.2005.03.003

Brewer, M. B. (1999). The psychology of prejudice: Ingroup love and outgroup hate?. *Journal of social issues, 55,* 429-444. https://doi.org/10.1111/0022-4537.00126

Brohan, E., Slade, M., Clement, S., & Thornicroft, G. (2010). Experiences of mental illness stigma, prejudice and discrimination: a review of measures. *BMC health services research, 10*, 80. https://doi.org/10.1186/1472-6963-10-80

Bukh, J. D., Bock, C., Vinberg, M., & Kessing, L. V. (2013). The effect of prolonged duration of untreated depression on antidepressant treatment outcome. *Journal of affective disorders, 145*, 42-48. https://doi.org/10.1016/j.jad.2012.07.008

Carter, A., Denton, A., Ladino, L. D., Hassan, I., Sawchuk, T., Snyder, T., ... & Group, P. C. (2018). Experience of psychogenic nonepileptic seizures in the Canadian league against epilepsy: A survey describing current practices by neurologists and epileptologists. *Seizure*, *61*, 227-233. https://doi.org/10.1016/j.seizure.2018.08.025

Carton, S., Thompson, P. J., & Duncan, J. S. (2003). Non-epileptic seizures: patients' understanding and reaction to the diagnosis and impact on outcome. *Seizure, 12*, 287-294. https://doi.org/10.1016/S1059-1311(02)00290-X

Chang, C. K., Hayes, R. D., Perera, G., Broadbent, M. T., Fernandes, A. C., Lee, W. E., ... & Stewart, R. (2011). Life expectancy at birth for people with serious mental illness and other major disorders from a secondary mental health care case register in London. *PloS one, 6*, e19590. doi: 10.1371/journal.pone.0019590

Clement, S., Schauman, O., Graham, T., Maggioni, F., Evans-Lacko, S., Bezborodovs, N., ... & Thornicroft, G. (2015). What is the impact of mental health-related stigma on help-seeking? A systematic review of quantitative and qualitative studies. *Psychological medicine*, *45*, 11-27. https://doi.org/10.1017/S0033291714000129

Corrigan, P. W., Druss, B. G., & Perlick, D. A. (2014). The impact of mental illness stigma on seeking and participating in mental health care. *Psychological Science in the Public Interest, 15*, 37-70. https://doi.org/10.1177/1529100614531398

Corrigan, P. W., & Wassel, A. (2008). Understanding and influencing the stigma of mental illness. *Journal of psychosocial nursing and mental health services, 4*, 2-48. https://doi.org/10.3928/02793695-20080101-04

Crocker, J., & Lutsky, N. (1986). Stigma and the dynamics of social cognition. In The dilemma of difference (pp. 95-121). Springer, Boston, MA.

David, A. S. (2012). Functional disorders, Cartesian dualism and stigma: where does the dualism really lie? 83, 869, http://dx.doi.org/10.1136/jnnp-2012-303051

Dell'Osso, B., & Altamura, A. C. (2010). Duration of untreated psychosis and duration of untreated illness: new vistas. *CNS spectrums, 15*, 238-246. https://doi.org/10.1016/j.neulet.2016.10.003

Devine, P. G., & Sharp, L. B. (2009). *Automaticity and control in stereotyping and prejudice. Handbook of prejudice, stereotyping, and discrimination.* Oxford, Oxfrod University press.

Duncan, R., Graham, C. D., & Oto, M. (2014). Outcome at 5–10 years in psychogenic nonepileptic seizures: what patients report vs. what family doctors report. *Epilepsy & behavior, 37,* 71-74. https://doi.org/10.1016/j.yebeh.2014.06.011

Dunne, A., Carolan, R., Swords, L., & Fortune, G. (2019). Patient and family perspectives of paediatric psychogenic non-epileptic seizures: A systematic review. Seizure.71, 279-285 https://doi.org/10.1016/j.seizure.2019.08.014

Durna, G., Yorulmaz, O., & Aktaç, A. (2019). Public stigma of obsessive compulsive disorder and schizophrenic disorder: Is there really any difference?. *Psychiatry research, 271,* 559-564. https://doi.org/10.1016/j.psychres.2018.12.065

du Toit, A., & Pretorius, C. (2017). Psychogenic nonepileptic seizures: Namibian healthcare providers' perceptions and frustrations. *Seizure, 50,* 43-52. https://doi.org/10.1016/j.seizure.2017.06.004

Earnshaw, V. A., & Quinn, D. M. (2012). The impact of stigma in healthcare on people living with chronic illnesses. *Journal of health psychology, 17,* 157-168. doi: 10.1177/1359105311414952

Eknoyan, G. (2006). A history of obesity, or how what was good became ugly and then bad. *Advances in chronic kidney disease, 13,* 421-427. https://doi.org/10.1053/j.ackd.2006.07.002

Fairclough, G., Fox, J., Mercer, G., Reuber, M., & Brown, R. J. (2014). Understanding the perceived treatment needs of patients with psychogenic nonepileptic seizures. *Epilepsy & behavior, 31,* 295-303. https://doi.org/10.1016/j.yebeh.2013.10.025

Francis, P., & Baker, G. A. (1999). Non-epileptic attack disorder (NEAD): a comprehensive review. *Seizure, 8,* 53-61. https://doi.org/10.1053/seiz.1998.0246

Fernando, S. (2006). Stigma, racism and power. *Aotearoa Ethnic Network Journal, 1,* 24-28.

Gigerenzer, G. (2008). Why heuristics work. *Perspectives on psychological science, 3,* 20-29. https://doi.org/10.1111/j.1745-6916.2008.00058.x

Glozier, N., Hough, C., Henderson, M., & Holland-Elliott, K. (2006). Attitudes of nursing staff towards co-workers returning from psychiatric and physical illnesses. *International Journal of Social Psychiatry, 52,* 525-534. https://doi.org/10.1017/S2045796017000178

Green, B., Norman, P., & Reuber, M. (2017). Attachment style, relationship quality, and psychological distress in patients with psychogenic non-epileptic seizures versus epilepsy. *Epilepsy & Behavior, 66*, 120-126. doi: 10.1016/j.yebeh.2016.10.015.

Kanemoto, K., LaFrance, W. C., Duncan, R., Gigineishvili, D., Park, S. P., Tadokoro, Y., ... & Kerr, M. (2017). PNES around the world: where we are now and how we can close the diagnosis and treatment gaps—an ILAE PNES Task Force report. *Epilepsia open, 2*, 307-316. ps://doi.org/10.1002/epi4.12060

Kibria, N. (2008). The 'new Islam'and Bangladeshi youth in Britain and the US. *Ethnic and racial studies, 31*, 243-266. https://doi.org/10.1080/01419870701337593

Nordentoft, M., Jeppesen, P., Petersen, L., Bertelsen, M., & Thorup, A. (2009). The rationale for early intervention in schizophrenia and related disorders. *Early intervention in psychiatry, 3*, S3-S7. doi: 10.1111/j.1751-7893.2009.00123.x

Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders (5th ed.)*. Washington, DC: Author.

Hamilton, D. L. (2015). Cognitive processes in stereotyping and intergroup behavior. New York: Psychology Press.

Henderson, C., Noblett, J., Parke, H., Clement, S., Caffrey, A., Gale-Grant, O., ... & Thornicroft, G. (2014). Mental health-related stigma in health care and mental health-care settings. *The Lancet Psychiatry, 1*, 467-482. https://doi.org/10.1016/S2215-0366(14)00023-6

Hightower, S., Carmon, M., & Minick, P. (2002). A qualitative descriptive study of the lived experiences of school-aged children with epilepsy. *Journal of pediatric health care, 16*, 131-137. https://doi.org/10.1067/mph.2002.119599

Hingray, C., El‐Hage, W., Duncan, R., Gigineishvili, D., Kanemoto, K., LaFrance Jr, W. C., ... & Wiseman, H. (2018). Access to diagnostic and therapeutic facilities for psychogenic nonepileptic seizures: an international survey by the ILAE PNES Task Force. *Epilepsia, 59*. 203-214. https://doi.org/10.1111/epi.13952

Hughto, J. M. W., Reisner, S. L., & Pachankis, J. E. (2015). Transgender stigma and health: A critical review of stigma determinants, mechanisms, and interventions. *Social science & medicine, 147*, 222-231. doi: 10.1016/j.socscimed.2015.11.010

Jones, S., Howard, L., & Thornicroft, G. (2008). 'Diagnostic overshadowing': worse physical health care for people with mental illness. *Acta Psychiatrica Scandinavica, 118*, 169-171. doi: 10.1111/j.1600-0447.2008.01211.x

Kanemoto, K., LaFrance, W. C., Duncan, R., Gigineishvili, D., Park, S. P., Tadokoro, Y., ... & Kerr, M. (2017). PNES around the world: where we are now and how we can close the diagnosis and treatment gaps—an ILAE PNES Task Force report. *Epilepsia open, 2*, 307-316. https://doi.org/10.1002/epi4.12060

Karterud, H. N., Knizek, B. L., & Nakken, K. O. (2010). Changing the diagnosis from epilepsy to PNES: patients' experiences and understanding of their new diagnosis. *Seizure, 19*, 40-46. https://doi.org/10.1016/j.seizure.2009.11.001

Karterud, H. N., Haavet, O. R., & Risør, M. B. (2016). Social participation in young people with nonepileptic seizures (NES): A qualitative study of managing legitimacy in

everyday life. *Epilepsy & Behavior*, *57*, 23-28. https://doi.org/10.1016/j.yebeh.2016.01.009

Lai, Y. M., Hong, C. P. H., & Chee, C. Y. (2001). Stigma of mental illness. *Singapore medical journal*, *42*(3), 111-114. https://doi.org/10.1067/mph.2002.119599

Lee, R. S., Kochman, A., & Sikkema, K. J. (2002). Internalized stigma among people living with HIV-AIDS. *AIDS and Behavior*, *6*, 309-319. https://doi.org/10.1023/A:1021144511957

Lepore, S. J., & Greenberg, M. A. (2002). Mending broken hearts: Effects of expressive writing on mood, cognitive processing, social adjustment and health following a relationship breakup. *Psychology and Health*, *17*, 547-560. https://doi.org/10.1192/apt.11.5.338

Link, B. G., & Phelan, J. C. (2001). Conceptualizing stigma. *Annual review of Sociology*, *27*, 363-385. https://doi.org/10.1146/annurev.soc.27.1.363

Livingston, J. D., & Boyd, J. E. (2010). Correlates and consequences of internalized stigma for people living with mental illness: A systematic review and meta-analysis. *Social science & medicine, 71*, 2150-2161. doi: 10.1016/j.socscimed.2010.09.030

Looper, K. J., & Kirmayer, L. J. (2004). Perceived stigma in functional somatic syndromes and comparable medical conditions. *Journal of psychosomatic research, 57*, 373-378. https://doi.org/10.1016/j.jpsychores.2004.03.005

Ludwig, L., Pasman, J. A., Nicholson, T., Aybek, S., David, A. S., Tuck, S., ... & Stone, J. (2018). Stressful life events and maltreatment in conversion (functional neurological) disorder: systematic review and meta-analysis of case-control studies. *The Lancet Psychiatry, 5*, 307-320. https://doi.org/10.1016/S2215-0366(18)30051-8

MacLeod, J. S., & Austin, J. K. (2003). Stigma in the lives of adolescents with epilepsy: a review of the literature. *Epilepsy & Behavior, 4,* 112-117. https://doi.org/10.1016/S1525-5050(03)00007-6

Macinnes, D.L., & Lewis, M. (2008). Evaluation of a short group programme to reduce self stigma in people with serious and enduring mental health problems. *Journal of Psychiatric and Mental Health Nursing, 15,* 59–65. https://doi.org/10.1111/j.1365-2850.2007.01212.x

MacLeod, J. S., & Austin, J. K. (2003). *Stigma in the lives of adolescents with epilepsy: a review of the literature. Epilepsy & Behavior, 4,* 112-117. https://doi.org/10.1016/S1525-5050(03)00007-6

Macrae, C. N., Milne, A. B., & Bodenhausen, G. V. (1994). Stereotypes as energy-saving devices: A peek inside the cognitive toolbox. *Journal of Personality and Social Psychology, 66,* 37-47. http://dx.doi.org/10.1037/0022-3514.66.1.37

McMillan, K. K., Pugh, M. J., Hamid, H., Salinsky, M., Pugh, J., Noël, P. H., ... & LaFrance Jr, W. C. (2014). Providers' perspectives on treating psychogenic nonepileptic seizures: frustration and hope. *Epilepsy & Behavior, 37,* 276-281. https://doi.org/10.1016/j.yebeh.2014.07.001

McNeil, K., Brna, P. M., & Gordon, K. E. (2012). Epilepsy in the Twitter era: a need to re-tweet the way we think about seizures. *Epilepsy & behavior, 23,* 127-130. https://doi.org/10.1016/j.yebeh.2011.10.020

McWilliams, A., Reilly, C., McFarlane, F. A., Booker, E., & Heyman, I. (2016). Nonepileptic seizures in the pediatric population: a qualitative study of patient and family experiences. *Epilepsy & Behavior, 59,* 128-136. https://doi.org/10.1016/j.yebeh.2016.03.029

Mellers, J. D. C. (2005). The approach to patients with “non-epileptic seizures”. *Postgraduate medical journal, 81*, 498-504. http://dx.doi.org/10.1136/pgmj.2004.029785

Millogo, A., Ngowi, A. H., Carabin, H., Ganaba, R., Da, A., & Preux, P. M. (2019). Knowledge, attitudes, and practices related to epilepsy in rural Burkina Faso. Epilepsy & Behavior, 95, 70-74. https://doi.org/10.1016/j.yebeh.2019.03.006

Mittal, D., Sullivan, G., Chekuri, L., Allee, E., & Corrigan, P. W. (2012). Empirical studies of self-stigma reduction strategies: a critical review of the literature. *Psychiatric Services, 63*, 974-981. doi: 10.1176/appi.ps.201100459

Mojtabai, R., Olfson, M., Sampson, N. A., Jin, R., Druss, B., Wang, P. S., ... & Kessler, R. C. (2011). Barriers to mental health treatment: results from the National Comorbidity Survey Replication. *Psychological medicine, 41*, 1751-1761. https://doi.org/10.1017/S0033291710002291

Pickett, K. E., James, O. W., & Wilkinson, R. G. (2006). Income inequality and the prevalence of mental illness: a preliminary international analysis. *Journal of Epidemiology & Community Health, 60*, 646-647. doi: 10.1136/jech.2006.046631

Pickett, K. E., & Wilkinson, R. G. (2010). Inequality: an underacknowledged source of mental illness and distress. *The British Journal of Psychiatry, 197*, 426-428. https://doi.org/10.1192/bjp.bp.109.072066

Pretorius, C. (2016). Barriers and facilitators to reaching a diagnosis of PNES from the patients’ perspective: preliminary findings. *Seizure, 38*, 1-6. https://doi.org/10.1016/j.seizure.2016.03.007

Pretorius, C., & Sparrow, M. (2015). Life after being diagnosed with psychogenic non-epileptic seizures (PNES): A South African perspective. *Seizure, 30,* 32-41. https://doi.org/10.1016/j.seizure.2015.05.008

Jacoby, A. (2008). Epilepsy and stigma: an update and critical review. *Current neurology and neuroscience reports, 8,* 339. https://doi.org/10.1007/s11910-008-0052-8

Jilek-Aall, L., Jilek, M., Kaaya, J., Mkombachepa, L., & Hillary, K. (1997). P*sychosocial study of epilepsy in Africa. Social science & medicine, 45,* 783-795. doi: 10.1016/s0277-9536(96)00414-5

Johnstone, L., & Boyle, M. (2018). The Power Threat Meaning Framework: An Alternative Nondiagnostic Conceptual System. Journal of Humanistic Psychology. https://doi.org/10.1177/0022167818793289

Jones, S., Howard, L., & Thornicroft, G. (2008). 'Diagnostic overshadowing': worse physical health care for people with mental illness. *Acta Psychiatrica Scandinavica, 118,* 169-171. doi: 10.1111/j.1600-0447.2008.01211.x

Rawlings, G. H., Brown, I., & Reuber, M. (2017). Deconstructing stigma in psychogenic nonepileptic seizures: an exploratory study. *Epilepsy & Behavior, 74,* 167-172. https://doi.org/10.1016/j.yebeh.2017.06.014

Rawlings, G. H., Brown, I., & Reuber, M. (2018). Narrative analysis of written accounts about living with epileptic or psychogenic nonepileptic seizures. *Seizure, 62,* 59-65. https://doi.org/10.1016/j.seizure.2018.09.022

Rawlings, G. H., Brown, I., Stone, B., & Reuber, M. (2018). Written accounts of living with epilepsy or psychogenic nonepileptic seizures: a thematic comparison. *Qualitative health research, 28,* 950-962. https://doi.org/10.1177/1049732317748897

Rawlings, G. H., Brown, I., Stone, B., & Reuber, M. (2018). A pilot randomised controlled trial of a home-based writing intervention for individuals with seizures. *Psychology & health, 33*, 1151-1171. https://doi.org/10.1080/08870446.2018.1478974

Reuber, M. (2008). Psychogenic nonepileptic seizures: answers and questions. *Epilepsy & behavior, 12*, 622-635. https://doi.org/10.1016/j.yebeh.2007.11.006

Reuber, M., Pukrop, R., Bauer, J., Derfuss, R., & Elger, C. E. (2004). Multidimensional assessment of personality in patients with psychogenic non-epileptic seizures. *Journal of Neurology, Neurosurgery & Psychiatry, 75*, 743-748. doi: 10.1136/jnnp.2003.013821

Reynaert, C. C., & Gelman, S. A. (2007). The influence of language form and conventional wording on judgments of illness. *Journal of Psycholinguistic Research, 36*, 273-295. doi: 10.1007/s10936-006-9045-4

Roberts, K. (2019). The Role of Positivity: Physical and Mental Aspects of Lower Limb Amputation Due to Diabetes. *Diabetic Medicine, 31*. doi: 10.1111/dme.12474

Robson, C., Myers, L., Pretorius, C., Lian, O. S., & Reuber, M. (2018). Health related quality of life of people with non-epileptic seizures: The role of socio-demographic characteristics and stigma. *Seizure, 55*, 93-99. https://doi.org/10.1016/j.seizure.2018.01.001

Robson, C., & Lian, O. S. (2017). "Blaming, shaming, humiliation": Stigmatising medical interactions among people with non-epileptic seizures. *Wellcome open research, 2*.

Rommelfanger, K. S., Factor, S. A., LaRoche, S., Rosen, P., Young, R., & Rapaport, M. H. (2017). Disentangling stigma from functional neurological disorders: conference

report and roadmap for the future. *Frontiers in neurology*, *8*, 106. https://doi.org/10.1016/j.jesp.2015.04.008

Thompson, A. R., Clarke, S. A., Newell, R., Gawkrodger, G., & The Appearance Research Collaboration. (2010). Vitiligo linked to stigmatisation in British South Asian women: A qualitative study of the experiences of living with vitiligo. *The British Journal of Dermatology,163,* 481-486. doi: 1 0.1111/j.1365-2133.2010.09828.x.

Tong, X., An, D., Reuber, M., Zhang, Q., & Zhou, D. (2018). Diagnostic and treatment services for patients with Psychogenic Nonepileptic Seizures (PNES): A survey of health professionals in urban China. *Seizure*, *56*, 78-87.

Thornicroft, G. (2011). Physical health disparities and mental illness: the scandal of premature mortality. *The British Journal of Psychiatry*, *199*, 441-442. doi: 10.1192/bjp.bp.111.092718

Weisel, O., & Böhm, R. (2015). "Ingroup love" and "outgroup hate" in intergroup conflict between natural groups. *Journal of experimental social psychology*, *60*, 110-120. https://doi.org/10.1016/j.jesp.2015.04.008

Whippy, N., & Lusher, J. (2019). A Systematic Review Examining Conflicting Attitudes Around Safety and Legislation Towards People Driving with Epilepsy. International *Journal of Psychology and Cognitive Science*, *5*, 88-94.

Wykes, T., & Hayward, P. (2006). Group treatment of perceived stigma and self‐esteem in schizophrenia: a waiting list trial of efficacy. *Behavioural and Cognitive Psychotherapy, 34,* 305–318. https://doi.org/10.1017/S1352465805002705

Vaidya-Mathur, U., Myers, L., Laban-Grant, O., Lancman, M., Lancman, M., & Jones, J. (2016). Socialization characteristics in patients with psychogenic nonepileptic seizures (PNES). *Epilepsy & Behavior, 56,* 59-65. https://doi.org/10.1016/j.yebeh.2015.12.032

Voon, V., Cavanna, A. E., Coburn, K., Sampson, S., Reeve, A., LaFrance Jr, W. C., & American Neuropsychiatric Association Committee for Research. (2016). Functional neuroanatomy and neurophysiology of functional neurological disorders (conversion disorder). *The Journal of neuropsychiatry and clinical neurosciences, 28,* 168-190. https://doi.org/10.1016/j.jpsychores.2016.10.007

Sabat, I. E., Lindsey, A. P., King, E. B., Winslow, C., Jones, K. P., Membere, A., & Smith, N. A. (2019). Stigma Expression Outcomes and Boundary Conditions: A Meta-Analysis. *Journal of Business and Psychology, 1-16.* https://doi.org/10.1007/s10869-018-9608-z

Shefer, G., Cross, S., Howard, L. M., Murray, J., Thornicroft, G., & Henderson, C. (2015). Improving the diagnosis of physical illness in patients with mental illness who present in emergency departments: consensus study. *Journal of psychosomatic research, 78,* 346-351. https://doi.org/10.1016/j.jpsychores.2015.01.010

Schomerus, G., & Angermeyer, M. C. (2008). Stigma and its impact on help-seeking for mental disorders: what do we know?. Epidemiology and Psychiatric Sciences, 17, 31-37. https://doi.org/10.1017/S1121189X00002669

Schulze, B. (2007). Stigma and mental health professionals: a review of the evidence on an intricate relationship. *International review of Psychiatry, 19,* 137-155. doi: 10.1080/09540260701278929

Schnyder, N., Panczak, R., Groth, N., & Schultze-Lutter, F. (2017). Association between mental health-related stigma and active help-seeking: systematic review and meta-analysis. *The British Journal of Psychiatry, 210,* 261-268. https://doi.org/10.1192/bjp.bp.116.189464

Sharac, J., Mccrone, P., Clement, S., & Thornicroft, G. (2010). The economic impact of mental health stigma and discrimination: a systematic review. *Epidemiology and Psychiatric Sciences, 19* 223-232.

Spencer, S. J., Fein, S., Wolfe, C. T., Fong, C., & Duinn, M. A. (1998). Automatic activation of stereotypes: The role of self-image threat. Personality and Social Psychology Bulletin, 24(11), 1139-1152. doi: 10.1017/s1121189x00001159

Smith, B. J. (2014). Closing the Major Gap in PNES Research: Major Gap in PNES Research. *Epilepsy Currents, 14,* 63–67. https://doi.org/10.5698/1535-7597-14.2.63

Smyth, J., & Helm, R. (2003). Focused expressive writing as self‐help for stress and trauma. *Journal of clinical psychology*, *59*, 227-235. https://doi.org/10.1002/jclp.10144

Takeda, M. B., Helms, M. M., & Romanova, N. (2006). Hair color stereotyping and CEO selection in the United Kingdom. *Journal of human behavior in the social environment, 13,* 85-99. doi: 10.1300/J137v13n03_06

Wissel, B. D., Dwivedi, A. K., Gaston, T. E., Rodriguez-Porcel, F. J., Aljaafari, D., Hopp, J. L., ... & Moseley, B. D. (2016). Which patients with epilepsy are at risk for psychogenic nonepileptic seizures (PNES)? A multicenter case–control study. *Epilepsy & Behavior, 61,* 180-184. doi: 10.1016/j.yebeh.2016.05.03

Worsely, C., Whitehead, K., Kandler, R., & Reuber, M. (2011). Illness perceptions of health care workers in relation to epileptic and psychogenic nonepileptic seizures. *Epilepsy & Behavior, 20*, 668-673. https://doi.org/10.1016/j.yebeh.2011.01.029

Wyatt, C., Laraway, A., & Weatherhead, S. (2014). The experience of adjusting to a diagnosis of non-epileptic attack disorder (NEAD) and the subsequent process of psychological therapy. *Seizure, 23*, 799-807. https://doi.org/10.1016/j.seizure.2014.06.012

Yanos, P. T., Lucksted, A., Drapalski, A. L., Roe, D., & Lysaker, P. (2015). Interventions targeting mental health self-stigma: A review and comparison. *Psychiatric rehabilitation journal, 38*, 171. doi: 10.1037/prj0000100

Yanos, P. T., Roe, D., West, M. L., Smith, S. M., & Lysaker, P. H. (2012). Group-based treatment for internalized stigma among persons with severe mental illness: Findings from a randomized controlled trial. *Psychological Services, 9*, 248. https://doi.org/10.1037/a0028048

Yogarajah, M., Child, R., Agrawal, N., Cope, S., Edwards, M., & Mula, M. (2019). Functional seizures: An evaluation of the attitudes of general practitioners local to a tertiary neuroscience service in London. *Epilepsia Open, 4,* 54-62. ttps://doi.org/10.1002/epi4.12283

Appendices

Appendix A – Full list of search terms

Search terms related to NEAD and stigma (OR used within columns)

NEAD	*And*	Stigmatization
Non-epileptic attack disorder		Stigmatisation
Non epileptic attack disorder		Stigma
PNES		Prejudice
psychogenic non-epileptic seizure		Discrimination
psychogenic non epileptic seizure		
non-epileptic attack		
non epileptic attack		
non-epileptic seizure		
non epileptic seizure		
psychogenic seizures		
functional seizures		
dissociative seizures		
pseudo-seizures		

Appendix B: Coding scheme

	Information extracted from final sample
1	Authors
2	Year of publication
3	Country of origin
4	Database found
5	Publication type
6	Study design
7	Exclusion and inclusion criteria
8	Sample size
9	Age (M, range where available)
10	Demographic information
11	Inclusion of healthy comparison group (if appropriate)
12	Inclusion of clinical comparison group (if appropriate)
13	Statistical methods used (if appropriate)
14	Qualitative method used (if appropriate)
15	Stigma measures used
16	Other measures used
17	Finding

Appendix C - Checklist for Qualitative Research

JBI Critical Appraisal Checklist for Qualitative Research

Reviewer_____Date_____

Author _____Year_____Record Number_____

		Yes	No	Unclear	Not applicable
1.	Is there congruity between the stated philosophical perspective and the research methodology?	☐	☐	☐	☐
2.	Is there congruity between the research methodology and the research question or objectives?	☐	☐	☐	☐
3.	Is there congruity between the research methodology and the methods used to collect data?	☐	☐	☐	☐
4.	Is there congruity between the research methodology and the representation and analysis of data?	☐	☐	☐	☐
5.	Is there congruity between the research methodology and the interpretation of results?	☐	☐	☐	☐
6.	Is there a statement locating the researcher culturally or theoretically?	☐	☐	☐	☐
7.	Is the influence of the researcher on the research, and vice- versa, addressed?	☐	☐	☐	☐
8.	Are participants, and their voices, adequately represented?	☐	☐	☐	☐
9.	Is the research ethical according to current criteria or, for recent studies, and is there evidence of ethical approval by an appropriate body?	☐	☐	☐	☐
10.	Do the conclusions drawn in the research report flow from the analysis, or interpretation, of the data?	☐	☐	☐	☐

Overall appraisal: Include ☐ Exclude ☐ Seek further info ☐

Comments (Including reason for exclusion)

_____

_____

_____

Appendix D - Checklist for Analytical Cross Sectional Studies

THE JOANNA BRIGGS INSTITUTE

JBI Critical Appraisal Checklist for Analytical Cross Sectional Studies

Reviewer_____Date_____

Author _____Year_____Record Number_____.

	Yes	No	Unclear	Not applicable
1. Were the criteria for inclusion in the sample clearly defined?	☐	☐	☐	☐
2. Were the study subjects and the setting described in detail?	☐	☐	☐	☐
3. Was the exposure measured in a valid and reliable way?	☐	☐	☐	☐
4. Were objective, standard criteria used for measurement of the condition?	☐	☐	☐	☐
5. Were confounding factors identified?	☐	☐	☐	☐
6. Were strategies to deal with confounding factors stated?	☐	☐	☐	☐
7. Were the outcomes measured in a valid and reliable way?	☐	☐	☐	☐
8. Was appropriate statistical analysis used?	☐	☐	☐	☐

Overall appraisal: Include ☐ Exclude ☐ Seek further info ☐

Comments (Including reason for exclusion)

_____

_____

_____

Appendix E - Randomized Controlled Trials

THE JOANNA BRIGGS INSTITUTE

JBI Critical Appraisal Checklist for Randomized Controlled Trials

Reviewer_____Date_____

Author _____Year_____Record Number_____

		Yes	No	Unclear	NA
1.	Was true randomization used for assignment of participants to treatment groups?	☐	☐	☐	☐
2.	Was allocation to treatment groups concealed?	☐	☐	☐	☐
3.	Were treatment groups similar at the baseline?	☐	☐	☐	☐
4.	Were participants blind to treatment assignment?	☐	☐	☐	☐
5.	Were those delivering treatment blind to treatment assignment?	☐	☐	☐	☐
6.	Were outcomes assessors blind to treatment assignment?	☐	☐	☐	☐
7.	Were treatment groups treated identically other than the intervention of interest?	☐	☐	☐	☐
8.	Was follow up complete and if not, were differences between groups in terms of their follow up adequately described and analyzed?	☐	☐	☐	☐
9.	Were participants analyzed in the groups to which they were randomized?	☐	☐	☐	☐
10.	Were outcomes measured in the same way for treatment groups?	☐	☐	☐	☐
11.	Were outcomes measured in a reliable way?	☐	☐	☐	☐
12.	Was appropriate statistical analysis used?	☐	☐	☐	☐
13.	Was the trial design appropriate, and any deviations from the standard RCT design (individual randomization, parallel groups) accounted for in the conduct and analysis of the trial?	☐	☐	☐	☐

Overall appraisal: Include ☐ Exclude ☐ Seek further info ☐

Comments (Including reason for exclusion)

_____

_____

_____

Appendix F - Quality appraisal of final sample of studies

Qualitative	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10
1	Y	Y	Y	Y	Y	N	N	Y	Y	Y
2	Y	Y	Y	Y	Y	N	N	Y	Y	Y
3	Y	Y	Y	Y	Y	N	N	Y	Y	Y
4	Y	Y	Y	Y	Y	Y	N	Y	Y	Y
5	Y	Y	Y	Y	Y	N	N	Y	Y	Y
6	Y	Y	Y	Y	Y	N	N	Y	Y	Y
7	Y	Y	Y	Y	Y	N	N	Y	Y	Y
8	Y	Y	Y	Y	Y	Y	N	Y	Y	Y
9	Y	Y	Y	Y	Y	N	N	Y	Y	Y
10	Y	Y	Y	Y	Y	N	N	Y	Y	Y
11	Y	Y	Y	Y	Y	N	N	Y	Y	Y
12	Y	Y	Y	Y	Y	N	N	Y	Y	Y
Cross sectional	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8		
13	Y	Y	N	Y	Y	Y	Y	Y		
14	Y	Y	N	Y	N	N	N	Y		
15	Y	Y	Y	Y	Y	N	Y	Y		
16	Y	Y	Y	Y	Y	N	Y	Y		
17	Y	Y	Y	Y	Y	N	Y	Y		
18	Y	Y	Y	Y	Y	N	Y	Y		
19	Y	Y	Y	Y	N	N	Y	Y		
20	Y	Y	Y	Y	Y	N	N	Y		
21	Y	Y	Y	Y	N	N	Y	Y		
RCT	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10	Q11	Q12	Q13
22	Y	Y	N	Y	N/A	N	Y	Y	Y	Y	Y	Y	Y

Part II: Research report

A cross-sectional study examine the relationship between Stigma and Self-Efficacy in individuals with epilepsy or nonepileptic attack disorder

Abstract

Objectives

This study aimed to investigate the relationship between perceived stigma and self-efficacy, depression, anxiety and symptom severity.

Method

120 individuals (NEAD = 68, Epilepsy = 52) were recruited from online support groups and in-person from a seizure clinic in the UK. Participants completed measures of perceived stigma, depression, anxiety and symptom severity.

Results

There were no differences in scores for participants recruited online or in-clinic. Participants with NEAD reported higher levels of perceived stigma (p=<0.05), depression (p=<0.01), anxiety (p=<0.05), and number of seizures experienced per year (p=<0.05), than those with epilepsy. Depression was above the clinical cut-off on the PHQ-9 for NEAD (M = 14.10) but not for Epilepsy (M = 9.32). Correlation analysis found that symptom severity was not correlated with perceived stigma for NEAD (r = .17) but it was for epilepsy (r=.43, p=<0.01). Multiple regression for both NEAD (p=<0.01) and epilepsy (p=<0.05), found that depression significantly predicted perceived stigma.

Conclusions

Results indicated that individuals with NEAD reported more perceived stigma than those with epilepsy and higher, clinical levels of depression. The severity of symptoms was not associated with perceived stigma for NEAD but it was with epilepsy. Depression was significant predictive factor for perceived stigma in both epilepsy and NEAD.

Practitioner points

- Individuals with NEAD report high levels of depression, above the clinical cut-off for the PHQ-9
- Individuals with NEAD report higher levels of perceived stigma than those with epilepsy.
- Depression was predictive of perceived stigma for both epilepsy and NEAD.

Limitations

- The cross-sectional nature of this study makes it hard to establish causality regarding stigma for NEAD and epilepsy.
- Given the different aetiologies of NEAD and epilepsy, comparing the different disorders may be a limitation.
- The depression measures (PHQ-9) may measure some aspects of the symptomology of NEAD and epilepsy, overestimating the levels of depression.

Introduction

Stigma can be defined as when a feature causes an individual and/or groups to be negatively differentiated from others based on a real or imagined characteristic (Goffman, 1962, 2009). Goffman, in his seminal work, posited that stigma can stop an individual and/or group from gaining full '…*social acceptance*'(pg.4). Evidence suggests that when individuals perceive that they are feeling stigmatised, there can be significant and long-term negative impacts upon their mental and physical health, as well as their prospective attainment (DeWall et al., 2010; Arslan, 2018; Connolly, 1989). The concept of stigma has been well-researched in the fields of physical and mental health, with evidence suggesting that individuals may experience stigma differently, by nature and degree, based on their specific disorder (Link & Phelan, 2001; Corrigan, Watson, & Barr, 2006; Taft et al., 2011).

Since Goffman's time the concept of stigmatisation has been elaborated and is not now considered to be a unitary concept. More recent work by Hatzenbuehler and Link (2014) suggests that stigma can be regarded as occurring at multiple levels; from the intra-personal (stigma towards one's self) to the interpersonal (stigmatization from a person towards a person) and finally to the structural-level (also known as institutional stigma; governmental policies and laws that are targeted at specific groups to cause social exclusion). The term 'enacted' stigma (also known as 'external' stigma) is used to describe stigma occurring at the interpersonal and/or structural level and is when and individual and/or group is treated pejoratively in a tangible way (Major & O'brien, 2005; Hatzenbuehler & Link, 2014). For example, being refused care because of religious beliefs or denied a job because of race. An example of enacted stigma at the structural level can be found in a study by Pachankis et al. (2015), who looked at the consequence of country level laws and policies designed to impede or restrict homosexual men. The study found that the homosexual men in European countries with higher levels of structural stigma towards homosexual men (for

example in Russia and Ukraine) had fewer sexual partners and reduced access to HIV-preventive services.

'Perceived' stigma (also known as 'internal' stigma) is a concept used to denote when an individual characterises the negative behaviour and actions of others to a specific characteristic of their self (Pryor, Reeder,Yeadon,& Hesson-McInnis, 2004). This can be a result of the internalisation of negative stereotypes and prejudices linked to the perceived stigmatising characteristic (Miller & Kaiser, 2001). A European cross-sectional study by Alonso (2009) demonstrated that individuals are less likely to seek support and care for mental health difficulties if their reported degree of perceived-stigma is higher.

Individuals feeling stigmatised can have serious consequences, such as reduced quality of life (Ross, 2017), lower rates of employment (Link, Castille, & Stuber, 2008) and less overall life satisfaction (Rosenfield, 1997). Given these consequences of perceived stigma, there has been much research looking at the links between perceived stigma and treatment outcomes in the fields of physical and mental health (Hatzenbuehler, Phelan, & Link, 2013). For example, several studies have found a relationship between seeking treatment and perceived stigmatisation in conditions ranging from substance use disorders to mental health For example; high levels of perceived stigma were associated with reduced reported quality of life in individuals with schizophrenia (Cooper, Campbell, Larance, Murnion, & Nielsen, 2018; Staring et al. 2009; Jacoby & Austin. 2002; Cataldo, Jahan, & Pongquan, 2012; Lillis, Levin, & Hayes, 2011)

Epilepsy is a relatively common neurological disorder with over 500,000 individuals estimated to be affected in the UK (Epilepsy Society, 2018). Epilepsy can be described as a disease of the brain, when an individual experiences at least two unprovoked or reflex[1]

[1] A reflex seizure is a seizure is in response to environmental sensory stimulation, for example strobe lighting

seizures that occur at least 24 hours apart or one unprovoked (or reflex) seizures. The likelihood of additional seizures increases to 60% after two unprovoked seizures (Fisher et al., 2014). A number of different factors (for example, brain lesions) can cause epilepsy but for up to two-thirds of individuals suffering with epilepsy, there may be no known cause (Cull & Goldstein, 2002). Individuals with epilepsy have no or very little control over when they experience seizures and the rate at which people can experience seizures can vary greatly. Epilepsy can be treated with both medication and life-style management, but about one third of individuals continue to experience seizures despite optimal management (Sirven, Pedley, & Wilterdink, 2018)

There is a longstanding association of epilepsy with stigma (Holmes, Bourke, & Plumpton, 2019). Historically, epilepsy has been linked to spirit or demonic possession and as well as being thought to be contagious (Yildirim, Ertem, Dirican, & Baybas, 2019). Indeed, research suggests that, in some parts of the world, epilepsy continues to be mistakenly believed to be caused by witchcraft or possession rather than the result of a neurological disorder (Baskind & Birbeck, 2005). Whilst there is evidence that societal attitudes towards epilepsy are becoming more positive in UK, there is also evidence that prejudices towards individuals with epilepsy continue to exist in the UK (Holmes, Bourke, & Plumpton, 2019). Furthermore, there is also evidence that individuals with epilepsy continue to experience stigma related to their disorder, particularly in lower-socioeconomic status countries (Newton, & Garcia, 2012).

Given the historical and widespread prejudices towards people with epilepsy, it is perhaps unsurprising that individuals with epilepsy perceive greater stigma than the general population (Jacoby, Snape, & Baker, 2005). These high levels of perceived stigma in individuals with epilepsy are associated with reduced quality of life. Indeed, in those with uncontrolled epilepsy levels of stigma are a better predictor of quality of life than seizure

frequency (McLaughlin, Pachana, & Mcfarland, 2008). This may be because individuals who report high levels of stigma experience a high degree of shame and guilt related to their epilepsy (Van Brakel, 2006) meaning that they have lower-levels of self-worth (Claesson, Birgegard, & Sohlberg, 2007), self-efficacy (Baldwin, Baldwin, & Ewald, 2006) and are less like to access social and/or medical support (de Souza & Salgado, 2006)

Non-epileptic Attack Disorder (NEAD) is a condition whereby an individual experiences attacks that are outwardly similar to epileptic seizures, but that are not associated with the neurobiological correlates of epilepsy (Benbadis, 2005). NEAD is a relatively common with an estimated 15 000 people with a diagnosis in the UK (Kanemoto, et al., 2017). Similar to epileptic seizures, most episodes of 'seizure' are not wilfully produced and typically result in a temporary disruption of normal functioning in visual, sensory, and cognitive domains. In the absence of specialist testing (involving video-electrographic, VEEG, recording of typical seizures) it can be difficult to differentiate between epileptic and non-epileptic seizures. As such, most individuals with NEAD initially receive an erroneous diagnosis of epilepsy, and experience invasive procedures and/or are prescribed potent antiepileptic medication before receiving an accurate diagnosis (Francis & Baker, 1999; LaFrance et al., 2013; Reuber et al., 2002).

Current research indicates that, like those with epilepsy, individuals with NEAD often experience a significant amount of perceived stigma (Karterud, Knizek, & Nakken, 2010; Rawlings & Reuber, 2016; Rawlings, Brown, Stone, & Reuber, 2017). Given the lack of biological correlates for NEAD, it is often regarded as a medically unexplained symptom (MUS: Oto et al., 2005) or Somatic Symptom Disorder. The current Diagnostic and statistical manual of mental disorders (American Psychiatric Association, 2013) criteria is:

"A) One or more somatic symptoms that are distressing or result in significant disruption of daily life.

B) Excessive thoughts, feelings, or behaviours related to the somatic symptoms or associated health concerns as manifested by at least one of the following:

1. Disproportionate and persistent thoughts about the seriousness of one's symptoms.

2. Persistently high level of anxiety about health or symptoms.

3. Excessive time and energy devoted to these symptoms or health concerns.

C) Although any one somatic symptom may not be continuously present, the state of being symptomatic is persistent (typically more than 6 months)."

MUS terminology is, however, controversial. Bansal and Burton (2019) argue that the term can be a distancing factor in the clinician/patient relationship and that it can trivialise and be dismissive of patient's individual experiences (see also; Greco, 2012). Marks and Hunter (2015) reported that only 15% of patients had positive attitudes towards the term MUS or Somatic Symptom Disorder. Consequently, the term '*persistent physical symptoms'* has been suggested as a more appropriate label. However, given that this term is not currently in common usage, MUS will continue to be used for the sake of clarity.

Thus most patients with NEAD fulfil the diagnostic criteria of a mental health disorder (American Psychiatric Association, 2013; World Health Organisation, 2011). There has been a significant amount of research looking at perceived stigma in individuals with a metal health disorder, in particular as a barrier to accessing treatment/support (Sickel, Seacat, & Nabors, 2014). There is evidence that the degree of reported stigma differs between different mental health disorders (for example; schizophrenia, anxiety and depression) is low

(Patten et al., 2016), meaning that most mental heath disorders report a similar degree of stigma.

A number of different possible explanations for the underlying causes of NEAD have been suggested, but recently the Integrative Cognitive Model (ICM: Brown, & Reuber, 2016; Reuber & Brown, 2017) has offered the most comprehensive and integrated explanation. The ICM (Brown, 2004) combines previous theories of dissociation, conversion, and somatization. Dissociation is a detachment from one's surroundings, often in reaction to intolerable emotional states, for example attempting to recall traumatic instances from one's past. Indeed, frequency of disassociation has been linked to individuals experiencing significant past traumas (Kienle et al 2017). Conversion is when an individual experiences repressed emotional distress as physical illness. Research indicates that many individuals who experience medically unexplained symptoms, also have experienced significant trauma in their past (Brown, 2004). Both dissociation and conversion underpin the modern theory of somatization, the idea that psychological distress can be experienced as physiological symptoms. The ICM combines and expands all three theories into an integrative model. It proposes that information (such as traumatic memories) stored in an individual's cognitive systems causes disruption to the interplay between conscious and preconscious information processing. This disruption is caused by an attempt to avoid and/or reduce the experience of emotions related to past trauma. The model then posits that symptom-focussed attention (hypervigilance for physical responses, catastrophization of identified symptoms, etc) results in the creation and maintenance of medically unexplained systems. Therefore the medically unexplained symptoms can be regarded as a form of maladaptive emotional regulation (Brown, 2004). The model further proposes that trauma is a significant factor in impeding standard internal self-regulatory processes (Wells and Matthews, 1994).

The tenet of the ICM, as it relates to NEAD, is that the individuals develop internal, preconscious hypotheses about how best to respond to internal or external cues. Some of these hypotheses may be maladaptive (termed ‘rogue representations’ by Brown (2004)) but the individuals’ internal cognitive systems might regard them as the best and most adaptive response or explanation to the stimuli at the time. An example of this might be an individual experiencing high levels of anxiety and their internal, preconscious system might regard it to be better to experience a seizure than continue experiencing this emotion (see fig. 1).

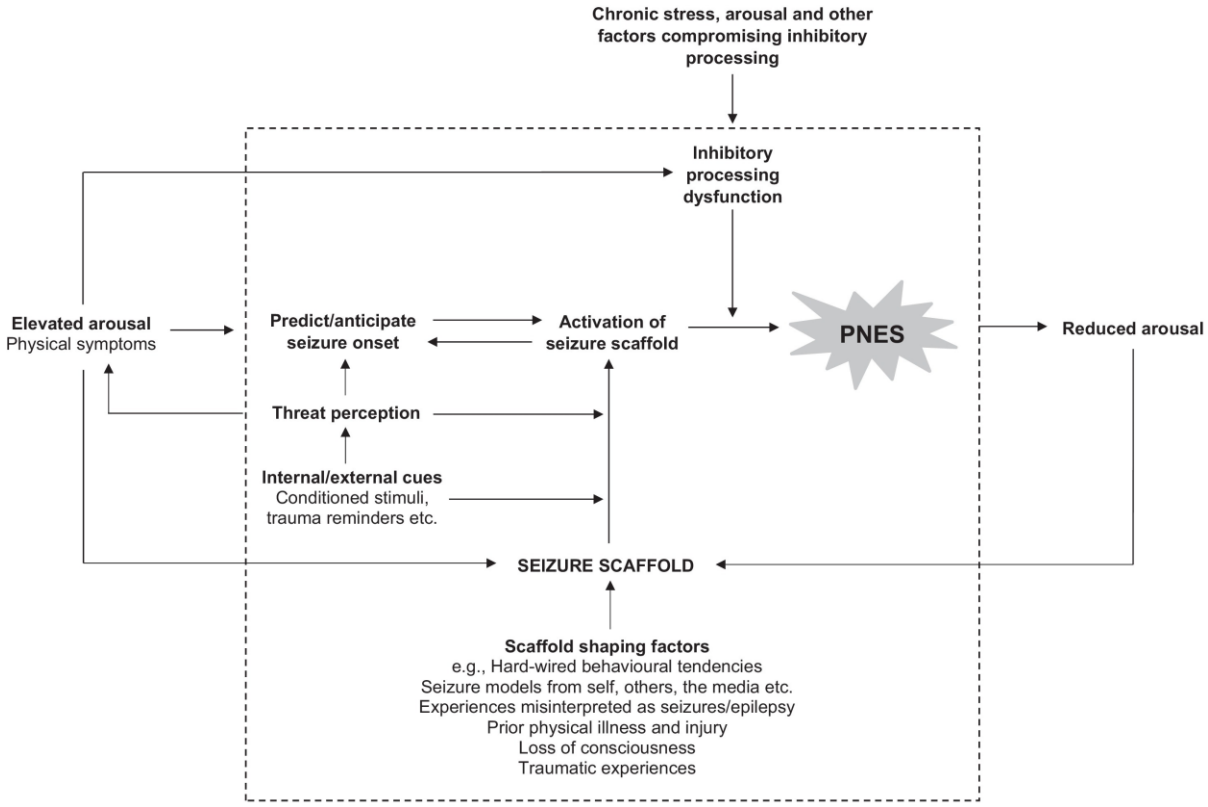


Fig. 1. The ICM of NEAD (Also known as Psychogenic non-epileptic seizures (PNES)). Important aspects are in the dashed area (from Reuber, Kanner, and Schachter, 2008).

There is a growing body of evidence for the ICM (Brown & Reuber, 2016). For example, a study by Garnefski, van Rood, De Roos, and Kraaij (2017), found that current somatic symptoms were significantly related to trauma-histories. Additionally, that these

history continue to cause strong and negative emotions in the present day. Furthermore, these somatic symptoms were significantly related to regular reported use of maladaptive cognitive coping strategies (e.g. self-blame, rumination, and catastrophisation) Additionally, a recent meta-analysis by Carlson and Perry (2017) found that 82% of individuals with NEAD who completed psychotherapy, reported a <50% reduction in seizures, supporting the cognitive-affective underlying cause. A study by Pick, Mellers, and Goldstein (2017) has indicated that individuals with NEAD report more past traumatic experiences than the general population Previous research has estimated the rates of a history of significant trauma in individuals with NEAD as being between 44-100% (Fiszman et al., 2004).

It has been suggested that one possible method for reducing the impact of trauma on the individuals is to increase their perception of their self-efficacy (Benight & Bandura, 2004). Self-efficacy is an individual's perception of their ability or capacity to engage in behaviours that will contribute toward them attaining their goals (Bandura, 2010). A systematic review of 27 papers (N=8011) by Luszczynska, Benight and Cieslak (2009) examining the relationship between self-efficacy and trauma found that self-efficacy had a medium to large effect on levels of distress, as well as on the frequency and severity of PTSD symptoms. Additionally, they found that increasing an individual's self-efficacy reduced misuse of substances and predicted fewer relapses. Samuelson, Bartel, Valadez and Jordan (2017), have also found that the self-efficacy moderates the relationship between traumatic events and the cognitive symptoms of PTSD. A study by DeCou, *et al.* (2019), looking at the relationship between negative public attitudes towards sexual assault, and psychological distress, found that high self-efficacy levels was a significant factor in ameliorating psychological distress.

An individual's perception of their self-efficacy is not stable, and it can both increase and decrease depending on situational factors (Madduz, 2016). Higher rates of reported self-

efficacy can also be developed through direct training (Eden & Aviram, 1993). High rates of self-efficacy have been indicated as positive treatment predictors in a number of different clinical populations, including substance misuse (Burleson & Kaminer, 2005), heart disease (Clark & Dodge, 1999), and bulimia (Bardone-Cone, 2006). These treatment predictors include treatment compliance (Mici et al., 2019) and post-treatment adherence to lifestyle changes at follow-up (Müller, Znoi & Moggi, 2019). Therefore, increasing self-efficacy may help to reduce psychological distress associated with past traumatic events and improve treatment compliance in NEAD populations.

Research has also established a link between perceived stigma and low self-efficacy. Kleim *et al*., (2008) found that higher perceived stigma scores in patients with schizophrenia were correlated with lower levels of self-efficacy regardless of symptom severity, insight, age and gender. Similar research by Landeen, Seeman, Goering, and Streiner, (2007), also found a correlation between perceived stigma and lower levels of self-efficacy in patients with schizophrenia. A study by Sung (2009), which took place in a Korean inpatient psychiatric hospital, also found a link between higher degrees of stigma and lower self-efficacy, and hypothesise that this relationship may act as a mediator for the low levels of quality of life found in the study. A systematic review and meta-analysis by Livingston and Boyd, (2010), examining stigma in people living with mental health problems, found that there was a strong negative relationship between degree of stigma and self-efficacy (among other psychosocial variables), which in turn negatively impacted treatment compliance.

As highlighted by Rawlings, Brown, and Reuber (2017) there is currently little available research examining the role of stigma in individuals with NEAD. The understanding of stigma in epilepsy is much further developed (De Boer, Mula, & Sander, 2008). There is also little current available research comparing stigma in NEAD and epilepsy groups, although as study conducted by Rawlings, Brown, and Reuber (2017) found that

individuals with NEAD were four-times more likely to report perceived stigma than individuals with epilepsy.

This study aims to investigate whether there are different reported levels of perceived stigma and self-efficacy in individuals with epilepsy and individuals with NEAD. There is little current literature looking at stigma in the NEAD population. Additionally, the study aims to understand how much of the variance within and between each population is accounted for by perceived self-efficacy.

Primary Aim and hypothesis

The primary aim of this study is to determine levels of perceived stigma and self-efficacy in patients with epilepsy or NEAD and to investigate the relationship between these factors and anxiety, depression, and seizure severity. The study specifically hypotheses that:

1) Participants with NEAD will report higher levels of self-rated perceived stigma than participants with Epilepsy.
2) Participants with NEAD will report lower levels of self-rated self-efficacy than participants with Epilepsy.
3) Self-efficacy will account for a greater level of variance in perceived stigma scores across both groups than levels of anxiety, depressive symptoms, and seizure severity.
4) Self-efficacy will account for a greater level of variance in perceived stigma scores than anxiety, depressive symptoms and seizure severity for participants with NEAD than participants with epilepsy

Method

Design

The current study was a quantitative, cross-sectional questionnaire study determining levels of stigma and self-efficacy and examining the amount of variance in perceived stigma

that can be accounted for by perceptions of self-efficacy between two groups: Individuals with epilepsy and individuals with NEAD.

Procedure

Participants were recruited in two ways. Firstly, via the outpatient seizure clinic at a large teaching hospital. Secondly via online advocacy groups. All participants who were recruited via the outpatient clinics were initially sent a participant information sheet (appendix A) with their routine appointment letter, approximately one-month before their appointment. They were then approached in the clinic and asked if they wished to participate in the study. Individuals who stated that they wished to participate were given a pack with the participant information sheet (appendix A), the consent form (appendix B) and study questionnaires (below). Participant's diagnoses were gleaned from their medical records following their signing of the consent form or from discussion with their neurologist if the diagnosis was unclear.

The second group were recruited via online advocacy groups (Epilepsy Action and FND action). The study was advertised between July-August 2019 on their main page and advertised via their Facebook and Twitter platforms. Participants were asked to complete an online consent (appendix B) form and then asked to complete the questionnaires (below). The online questionnaire was hosted on Qualtrics. Online participants gave self-reported diagnosis to either NEAD or Epilepsy. They were asked: "*Please provide you diagnosis (e.g. epilepsy, NEAD, Mixed epilepsy and NEAD etc):"*Demographic data regarding age and gender was also collected for both groups (appendix J)

Ethics

Ethical approval was granted by the Yorkshire and Humber NHS Ethics Committee (appendix C)

Participants

In order to be eligible to participate potential participants needed to be aged 16 or over and self-identify as having received a diagnosis of either epilepsy or NEAD. Individuals who reported they had a diagnosis of both were not eligible to be included.

Information about the study aims, procedure, right to withdraw, how the data would be stored, potential risks and options for further support were sent via letter to the participants recruited in the outpatient clinic or were present on the within the survey for the group recruited online (appendix A). Informed consent was gained by completion of the Informed consent sheet (appendix B). All personal and identifiable information were kept on an encrypted password protected database accessible by the primary investigator.

Questionnaires

The survey included the following questionnaires:

Stigma Scale for Chronic Illness (SSCI-8: Molina, Choi, Cella & Rao, 2013) (appendix D). The SSCI-8 is an eight-item questionnaire that measures internal stigma in individuals with neurological conditions (e.g. epilepsy, multiple sclerosis etc.). The reliability of the eight-item version of the SSCI has been demonstrated by Cronbach's alpha exceeding 0.7 (Cronbach's alpha = 0.89) and the validity by exceeding Cohen's Kappa of 0.40 with self-reported psychological distress (Cohen's Kappa = 0.42.7, SD = 19.7) (Rao et al., 2009).

The General Self-Efficacy Scale (GSE: Schwarzer & Jerusalem, 1995) (Appendix E). The GSE is a 10-item scale designed to assess an individual's perception of their ability to demonstrate personal mastery. The reliability of the GSE has been established by various studies with Cronbach's alpha ranging from .76 - .90 (Scholz, Gutiérrez-Doña, Sud, & Schwarzer, 2002).

Generalised Anxiety Disorder-7 (GAD-7:Lowe et al., 2008) (Appendix F). The GAD-7 is a 7-item measure of anxiety that is used in various clinical settings. Scores of 5, 10, and 15 are considered cut-offs for mild, moderate and severe anxiety with a score of 10 often regarded as the individual requiring further evaluation when used as a screening tool (Plummer, Manea, Trepel, & McMillan, 2016). Reliability of the GAD-7 has been demonstrated by the Cronbach's alpha exceeding 0.7 (Cronbach's alpha= 0.89).

Patient Health-Questionairre-9 (PHQ-9: Kroenke, Spitzer, & Williams, 2002) (Appendix G). The PHQ-9 is a widely used 9-item measure of depression. A cut-off score of 10 or greater (indicating symptoms of depression) produced a sensitivity of 89% (95% CI 0.83–0.92) and specificity of 0.85 (95% CI 0.75–0.91) (Manea, Gilbody, & McMillan, 2012) for depressive Symptoms. Estimates of internal reliability range from 0.86 to 0.89 using Cronbach's alpha (Kroenke, Spitzer & Williams, 2002). Test-retest reliability is estimated to be 0.84 with almost identical mean total scores.

Liverpool Seizure Severity Scale – Revised (LSSS-3: Scott-Lennox, Bryant-Comstock, Lennox, & Baker, 2001) (appendix H).The LSSS-3 is a 12-item scale that aims to assess the severity of an individual's seizure symptoms. The LSSS has been widely used in the epilepsy population. There is currently no current scale available for individuals with NEAD but the LSSS-3 has been used in NEAD populations previously (Green, Norman, & Reuber, 2017). Therefore, the LSSS-3 was used to assess seizure severity in the NEAD group. The LSSS-3 has demonstrated good reliability with a Cronbach's alpha of between 0.78-0.87, depending upon the participant group. The validity of the LSSS-3 was demonstrated by showing a correlation between change scores on the LSSS-3 and clinician judgement. This was shown to be significant at the 0.05 statistical significance level.

Power Analysis

G*Power3 was used to determine the sample size required to prevent type II errors. An a priori power analysis for linear regression fixed model was used with an effect size of F2= 0.15 (Faul, Erdfelder, Buchner, & Lang, 2009), an alpha of 0.05 and power of 0.80 (Bosco et al. 2015). This provides an overall sample size of 85. This has also been checked using Cohen's (1992) table and resulted in a similar sample size (84). A comparable study (Green, Norman, & Reuber, 2017) has been published and they were able to recruit 95 participants.

Analysis Plan

All the data was analysed using SPSS IBM Corp version 26 (2017). To investigate if there were different degrees of reported stigma between the two groups, independent sample t-tests were used to compare participants with epilepsy and participants with NEAD reports of perceived stigma. Participant groups were the independent variables and the score on the SSCI-8 was the dependent variable.

To understand if there were differences in reporting of self-efficacy between the two groups, independent sample t-test was used to compare participants with epilepsy and participants with NEAD for reported levels of self-efficacy. Participant groups were the independent variables and the score on the GSE were the dependent variable.

Multiple regression analysis was used to investigate if self-efficacy (dependent variable) accounts for variance on the perceived stigma scores (independent variable) across both groups once the other dependent variables (levels of anxiety, depressive symptoms and seizure severity) have been accounted for.

Results

Participant flow

Figure 1 shows the recruitment for online participants and figure 2 show the recruitment for clinic participants.

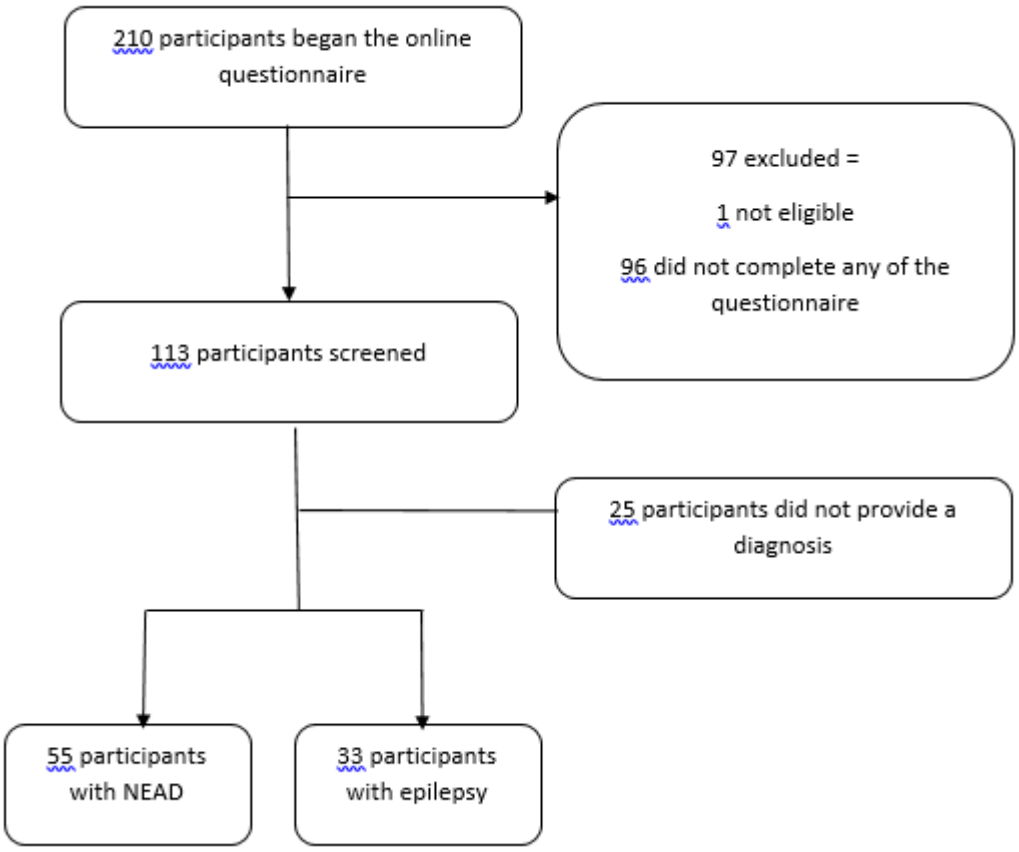


Figure 1. Online participant flow

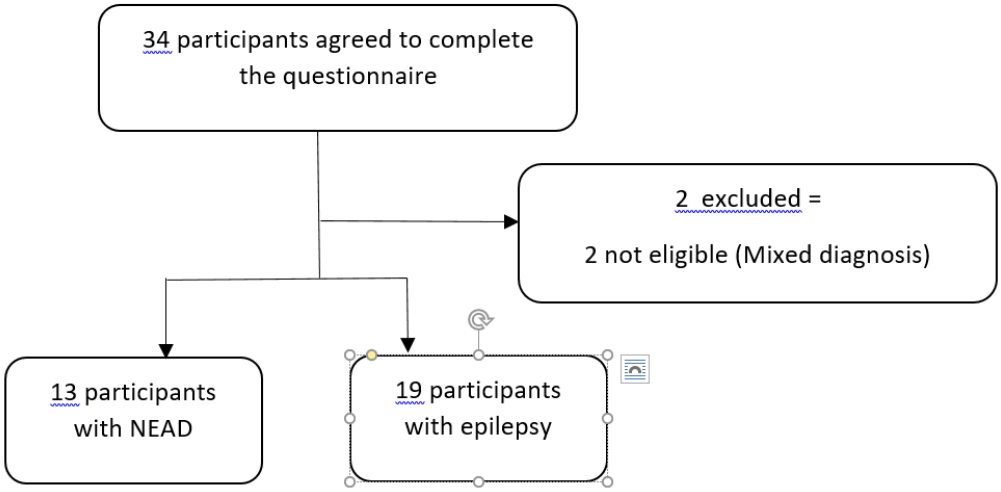


Figure 2. Clinic participant flow

Descriptive Statistics

A breakdown of the means and standard deviations for the different participant groups can be found in table 1 and 2. No significant differences were found on any of the self-report measures between online and clinic recruits in the epilepsy or NEAD groups. Participant data in the two clinical categories were combined for further analyses regardless of the source of recruitment (see tables 1 and table 2).

	NEAD		
	Online(*M* , *SD*)	Clinic(M , SD)	Independent t-tests
PHQ 9	14.47, 7.97	12.54, 7.75	.79
GAD 7	10.64, 6.75	11, 6.18	-.17
SSCI	24.64, 7.50	21.31, 9.43	1.37
GSE	25.73, 6.96	24.46, 5.56	.61
LSSS	19.25, 8.68	23.40, 6.07	-1.44
No. seizures per year	429.75, 766.971	272, 383.53	.53

Table 1: Participant means and standard deviations for NEAD participants in the online and clinic groups.

	Epilepsy		
	Online	Clinic	Independent t-tests
PHQ 9	10.12, 6.62	7.89, 6.163	1.17
GAD 7	7.30, 6.28	9.06, 8.03	-.84
SSCI	19.72, 950	15.67, 5.51	1.66
GSE	26.66, 9.17	26.79, 7.61	-.05
LSSS	18.80, 10.02	15.24, 9.03	1.21
No. seizures per year	127, 358	109, 162.99	.18

Table 2: Participant means and standard deviations for Epilepsy participants in the online and clinic groups.

Independent samples t-test

To investigate if participants with NEAD will report higher levels of self-rated perceived stigma than participants with Epilepsy., independent sample t-tests were calculated. There was a significant difference ($t(116)= 3.78, p=<0.01$) between the scores for participants with epilepsy (n=55, M= 18.26 , SD= 8.46) and NEAD (n=44, M=24.15, SD= 7.94) . The effect size for this analysis (d= .66) exceeded Cohen's (1988) convention for a medium effect size. These results indicate that participants with NEAD experience greater levels of perceived stigma than participants with epilepsy. There was no difference between participants with NEAD who were recruited online (n=55, M= 24.64, SD=7.50) and in clinic (n=13, M=21.31, SD=9.42) ($t(66)=1.37$,$p=.18$). Similarly, there no difference between participants with epilepsy who were recruited online (n=32, M=19.72, SD=9.50) and in clinic (n=18, M=15.67, SD=5.51) ($t(48)=1.66$, $p=.104$).

To investigate if participants with NEAD will report lower levels of self-rated self-efficacy than participants with epilepsy, independent sample t-tests were calculated. There was no significant difference in scores for participants with epilepsy (M= 25.49, SD=6.7) and NEAD (M = 26.71, 8.55 SD=6.69). This indicates that participants with both NEAD and epilepsy both report roughly comparable levels of self-efficacy.

The mean score for the PHQ-9 and GAD-7 scales was above the clinical cut-off point for NEAD but not epilepsy. 41 participants with NEAD scored above the clinical cut-off on the PHQ-9 compared to 23 with epilepsy. 38 participants with NEAD scored above the clinical cut-off for the GAD-7 compared to 27 for participants with Epilepsy.

	NEAD Online + Clinic (*M, SD*)	Epilepsy Online + Clinic (*M, SD*)	Independent t-tests	Effect size (Cohen's *d*)
PHQ 9	14.10, 7.91	9.32, 6.49	3.48**	0.66†
GAD 7	10.70, 6.61	7.88, 6.86	2.24*	0.41†
SSCI	24.15, 7.94	18.26, 8.46	3.78*	0.70†
GSE	25.49, 6.7	26.71, 8.55	-.87	-
LSSS	19.89, 8.43	17.51, 9.73	1.38	-
No. seizures per year	404.73, 719.79	120.62, 300.76	2.42*	0.51†

*p<.05, **p<.01, †Medium effect size (Cohen,1988)

Table 3: Comparing NEAD and Epilepsy using Independent samples t-tests.

Multiple Linear Regression

Correlation for participants with NEAD found that depression scores were correlated with anxiety, and perceived stigma and negatively correlated with self-efficacy. The analysis also found that self-efficacy was negatively correlated with anxiety and perceived stigma. Perceived stigma was correlated with anxiety (see table 4). For participants with Epilepsy, depression was found to be correlated with anxiety, and stigma and negatively correlated with self-efficacy. Self-efficacy was also negatively correlated with anxiety. Perceived stigma was correlated with depression, anxiety and symptom severity.

Correlations for NEAD participants

	PHQ-9	GAD-7	GSE	SSCI	LSSS	No. of seizures per year
PHQ-9	1	.83**	-.55**	.64**	.14	.08
GAD-7	.83**	1	-.50**	.56**	.16	.15
GSE	-.55**	-.50**	1	-.41**	-.16	.07
SSCI	.64**	.56**	-.41**	1	.17	.10
LSSS	.14	.16	-.16	.17	1	.02
No. Seizures per year	.08	.15	.07	.10	.02	1

**p <.01.

Table 4: Correlation analysis for participants with NEAD

Correlations for Epilepsy participants

	PHQ-9	GAD-7	GSE	SSCI	LSSS	No. of seizures per year
PHQ-9	1	.63**	-.32*	.58**	.28	.03
GAD-7	.63**	1	-.40**	.33*	.15	-.01
GSE	-.32*	-.40**	1	.06	.04	.04
SSCI	.58**	.33*	.06	1	.43**	-.08
LSSS	.28	.15	-.04	.43**	1	-.10
No. Seizures per year	.03	-.01	.04	-.08	-.10	1

* p<.05, **p <.01.

Table 5: Correlation analysis for participants with Epilepsy

To investigate if self-efficacy explains variance in perceived stigma scores above any variance explained by levels of anxiety, depressive symptoms, and seizure severity, two

multiple linear regression analysis was calculated for participant's with NEAD and epilepsy. The data met the assumptions necessary for the use of regression analysis (appendix K).

NEAD

A multiple regression was carried out to investigate whether the independent variables could significantly predict participants' with NEAD stigma scores. The results of the regression indicated that the model explained 36% of the variance and that the model was a significant predictor of stigma scores, (F (5,50) = 7.78, p< .000, R^2 $_{adjusted}$ = .36). While depression scores contributed significantly to the model (β = .423, t(55) = 2.179,p= .<03), anxiety scores (β = .136, t(55) = .72 ,p= .47), self-efficacy scores (β = -.14, t(55) = -1.09 ,p= .28), seizure severity scores (β = .04, t(55) = .37, p= .71) and number of reported seizures per year (β = .05, t(55) = .37, p= .65) did not significantly contribute towards the model.

Epilepsy

A multiple regression was carried out to investigate whether the independent variables could significantly predict participants' with NEAD stigma scores. The results of the regression indicated that the model explained 42% of the variance and that the model was a significant predictor of stigma scores (F (5,34)= 7.19, *p*<.000), R^2 $_{adjusted}$ = .443). Again, depression scores contributed significantly to the model (β = .57, t(34) = 3.56, p= .001) scores on anxiety (β = -.09, t(34) = -.6 ,p= .55), general self-efficacy (β = -.17, *t*(34) = -.599, *p*= .26), seizure severity scores (β = .26, t(34) = 1.87, p= .08) and number of reported seizures per year (β = -.05, *t*(34) = -.43, *p*= .67) did not significantly contribute towards the model.

Discussion

The results from this study suggest that depression may be predictive of perceived stigma for NEAD and epilepsy. This is perhaps not surprising given the available literature

linking depression and perceived stigma (Manos, Rusch, Kanter, & Clifford, 2009) and similar findings have been found for participants with epilepsy by Rawlings, Brown, and Reuber (2017). Manos, Rusch, Kanter, and Clifford (2009) suggest a model that may help explain the link between depression and perceived stigma. Although their focus is on individuals whose primary diagnosis is major depressive disorder, they argue that the symptoms of depression lead to an increase in the salience of stigmatising attitudes. For example, an attempt to hide seizures may lead to the avoidance of social situations, which in turn may increase the depressive feelings and perceived stigma (Ottenbreit & Dobson, 2004). Research has also indicated that the feeling of stigma can be predictive of depressive symptoms (Griffiths, Christensen, & Jorm, 2008; Livingston, & Boyd, 2010).

This is further supported by the finding that NEAD was associated with clinical levels of anxiety and depression, whereas epilepsy was associated with sub-clinical depression and anxiety. Evidence suggests that individuals who are depressed are more likely to appraise situations and social interactions negatively and show an impaired ability to recognise happiness in others (Joormann, & Gotlib, 2006; Leppänen, 2006; Surguladze, et al., 2004). This links to research which suggests the individuals with NEAD often report negative experiences of care from health professionals (Rawlings & Reuber, 2016; Rawlings, & Reuber, 2018) and that evidence suggests that there may be differences in interactions between individuals with NEAD and healthcare professionals compared with similar interactions between those with epilepsy and healthcare professionals (Monzoni, Duncan, Grünewald, & Reuber. (2011). Therefore, individuals with NEAD may be experiencing depression, which may make them more acutely aware of perceived stigma from others and more likely to regard interactions with others in a negative light. This may then be compounded by individuals with NEAD experiencing enacted stigma from others.

The link between depression and perceived stigma in individuals with epilepsy is supported by existing research (Rawlings, Brown, and Reuber, 2017). It is interesting that individuals with NEAD reported higher levels of depression than individuals with epilepsy and that the NEAD populations mean scores were above the clinical cut-off whilst the epilepsy participants were not. Nevertheless, much of the proposed model by Manos, Rusch, Kanter, and Clifford (2009) may also apply to participants with epilepsy; depression increases the salience of stigmatising experiences.

The higher depression and anxiety scores in NEAD also fits with the ICM (Brown, & Reuber, 2016; Reuber, & Brown, 2017). One possible explanation for NEAD suggested by the ICM is that the preconscious develops maladaptive responses (such as seizures) in response to internal or external stimuli. Seizures in NEAD might be regarded as a maladaptive form of emotional regulation. Therefore, higher levels of negatively experienced emotions would be expected in NEAD. It may also explain why symptom severity is not correlated with stigma in NEAD. The symptoms of NEAD may be easier to experience than intolerable emotional states that the seizures have been developed to avoid.

The finding from this study differ slightly from previous research in that depression and anxiety were correlated with perceived stigma for NEAD and epilepsy. In the study by Rawlings, Brown, and Reuber (2017), they found that depression and anxiety were correlated with perceived stigma for participants with epilepsy but not NEAD. It should be noted that whilst this study used the same measure for anxiety (GAD-7) as the current study, a different measure of depression was used. Rawlings, Brown and Reuber used the Neurological Disorders Depression Inventory for Epilepsy (NDDI-E; Oliveira, et al., 2011), as this study used the PHQ-7. The regression analysis indicated that depression scores were predictive of perceived stigma scores. Difficulties with depression have been found to be a significant problem for participants with epilepsy (e.g. Mula, 2017; Altın, 2018) and NEAD (e.g.

Szaflarski & Szaflarski, 2004; LaFrance & Syc, 2009) in the current research literature. There is also a well-established link between depression and perceived stigma (Pyne etal., 2004; Kanter, Rusch & Brondino, 2008; Manos, Rusch, Kanter, & Clifford, 2009).

Whilst there was no significant difference in symptom severity scores, NEAD was associated with higher reported seizures than participants with epilepsy. However, for epilepsy, symptom severity was correlated with perceived stigma, indicating that the symptom severity was related to perceived stigma in epilepsy. This is despite there not being a meaningful difference in symptom severity between the two groups. There was a significant difference in number of seizures experienced per year, with NEAD experiencing, on average, almost twice as many seizures as epilepsy. Therefore, despite experiencing more seizures and similar levels of symptom severity to epilepsy, the symptoms of NEAD were not linked to perceived stigma. This suggests that there might be different mechanisms for the development and perpetuation of perceived stigma in NEAD and epilepsy.

As expected, NEAD was associated with higher levels of perceived stigma than epilepsy. It is possible that the diagnosis of NEAD being regarded as a mental health disorder may be contributing to the high levels of reported perceived stigma, independent of symptom severity. There is a significant literature reporting on the stigma faced by those with mental health difficulties, at the intrapersonal (Corrigan, Watson, & Barr, 2006), interpersonal Couture, & Penn, 2003) and structural level (Corrigan, Markowitz, & Watson, 2004). As highlighted by Corrigan (2007), the diagnosis and label of mental illness may result in the person experiencing enacted stigma from professionals and the public, particularly in the form of stereotypes that perpetuate myths such as the homogeneity of mental illness and the lack of possible recovery. These arguments have been exemplified in research by Teachman, Wilson, and Komarovskaya, (2006) who found negative implicit attitudes (using the Implicit Attitude Test) regarding the helplessness and blameworthiness of individuals with mental

health difficulties and explicit negative attitudes regarding helplessness. Therefore, those who have been diagnosed with NEAD may have had implicit and explicit negative attitudes towards mental illnesses before their diagnosis, which may have contributed to the development of perceived stigma. It might also explain why symptom severity was not correlated with perceived stigma in NEAD, but it was in epilepsy; the stigma might be more related to the diagnosis rather than the symptoms. Teachman, Wilson, and Komarovskaya also found that these negative attitudes towards mental illness was not reduced in individuals with mental health problems, which may perpetuate the experiences of perceived stigma by NEAD.

The fact that participants with NEAD reported greater degrees of perceived stigma than participants with epilepsy, is similar to previous research in this area (Rawlings, Brown, & Reuber, 2017). The research on stigma in MUS is more mixed. Research by Taft et al. (2011) indicated that participants with irritable bowel syndrome (IBS; MUS) showed greater levels of perceived stigma than participants with Inflammatory Bowel Disorder (IBD: non-MUS). In contrast, research by Looper and Kirmayer (2004) indicated that whilst chronic fatigue syndrome (CFS) participants showed a greater degree of stigma compared to a matched medical condition with a clearer aetiology, participants with fibromyalgia (FM), or irritable bowel syndrome (IBS) showed no significant difference in perceived stigma scores to a matched medical condition. They argue that CFS has a greater medical ambiguity than FM or IBS. This may be similar for NEAD, which may, alongside the link to mental illness, explain the greater degree of perceived stigma.

Some existing research suggests that for some physical disorders, symptom severity has been linked to perceived stigma. For example, Taft et al. (2009) found that participants with IBD who reported a higher degree of symptom severity, also reported greater perceived stigma. Similar results have been found for participants with depression (Pyne et al., 2004)

and psoriasis (Böhm et al., 2014). Whilst this was not the case for participants with NEAD, it was for participants with epilepsy in the current study, where there was a correlation between perceived stigma and symptom severity. This contrasts with the work of Rawlings, Brown, and Reuber (2017) who found that symptom severity was not correlated with perceived stigma in epilepsy. However they did find that seizure frequency was correlated with the sequelae of epilepsy, such as memory and concentration difficulties. It is possible that the highly visible nature of epileptic seizures may result in similar emotional responses to those who experience IBD and psoriasis. Symptom related shame is a well-researched concept in all three disorders and may be a common factor in symptom related stigma (Trindade, Ferreira, & Pinto-Gouveia, 2017; Sampogna, Tabolli, & Abeni, 2012; Jacoby, & Austin, 2007).

Both NEAD and epilepsy were associated with similar levels of self-efficacy. This indicates that both groups felt themselves to be equally self-efficacious. There is little existing research looking at self-efficacy and perceived stigma in NEAD populations. However, there is existing research establishing a link between perceived stigma and self-efficacy in participants with schizophrenia (Kleim,et al.2008), bipolar and depression (Brohan et al., 2011), alcohol abuse (Schomerus, et al. 2011), and gambling addiction (Hing, Nuske, Gainsbury, & Russell, 2016). Research by DiIorio et al. (2003) also found an association between perceived stigma and self-efficacy for participants with epilepsy. The correlation analysis suggests that there was not a link between self-efficacy and perceived stigma for participants with epilepsy, but there was for participants with NEAD. However, the regression analysis indicated that self-efficacy did not account for a significant degree of the variance in perceived stigma for either group. One possible explanation for this is the high degree to which depression and self-efficacy were negatively correlated in NEAD, but less so in epilepsy. It is possible that the depression measure (PHQ-9) was tapping into similar (but

opposite) processes as the self-efficacy measure (GSE) in NEAD. The high correlation between depression and perceived stigma in NEAD may have also been capturing lower self-efficacy; explaining the high correlation between self-efficacy and perceived stigma, but the low explained variance of self-efficacy in perceived stigma.

There was no difference for participants recruited in clinic or online for either NEAD or epilepsy groups. This is perhaps surprising given that symptom severity might be expected to be higher in individuals who regularly attend clinics, but it cannot be ruled out that online participants were also regularly attending clinics.

Strengths, Limitations, and future directions

A major strength of this study is that it is, to our knowledge, the first study investigating the role of self-efficacy in perceived stigma in a NEAD and epilepsy population. As such, it contributes to the current literature on stigma and self-efficacy in NEAD. This study builds on the work of Rawlings, Brown, and Reuber (2017) in developing our understanding of the difference in stigma between NEAD and epilepsy.

The finding from this study should be considered in light of a number of limitations. Firstly, the cross-sectional nature of the study makes it difficult to establish causality. A longitudinal study, specifically examining pre/post diagnosis may allow a better untangling of the nature of perceived stigma in NEAD. Additionally, all the data was self-reported, which could be regarded as a weakness of the study. It was not possible to confirm the diagnosis of the online participants, meaning that they may not have had a formal diagnosis or may have a mixed diagnosis of epilepsy and NEAD. Additionally, the participants were all drawn from the UK, meaning that it may be problematic to generalise these findings to NEAD and epilepsy populations in other parts of the world.

Furthermore, comparing epilepsy and NEAD populations might be a limitation of this study. The aetiology of epilepsy and NEAD is very different, despite the outwardly similar appearance of the seizures. A comparison with other functional or psychological disorders might be a more appropriate comparison, especially to establish the level of perceived stigma experienced in NEAD in comparison to similar disorders. The different aetiologies may also have meant that some questions used were not the most appropriate. For example; it is possible that that the SSCI taps into different processes for epileptic and NEAD participants. The link between diagnosis of mental illness and stigma is well established and therefore the SSCI may be measuring the stigma associated with mental illness rather than specifically stigma related to non-epileptic attacks (Rüsch, Angermeyer, & Corrigan, 2005). This may also explain why there was not a link between symptom severity and perceived stigma in the NEAD group but there was for the epilepsy group. The SSCI focuses on the perception of the relationship with others. As highlighted by Rawlings, Brown, and Reuber (2017), it may be that when measuring perceived stigma in participants with NEAD, some of the results may also be measuring difficulties with interpersonal relationships. Green, Norman, and Reuber (2017), report high levels of attachment difficulties in individuals with NEAD, therefore the SSCI may also be reporting interpersonal difficulties for participants with NEAD, rather than just perceived stigma.

One possible explanation for the relatively high PHQ-9 scores in participants with epilepsy and the link between PHQ-9 scores and perceived stigma, is that some items of the PHQ-9 may be measuring symptoms of epilepsy, rather than symptoms of depression. Although the PHQ-9 has been validated for individuals with epilepsy (Rathore, 2014; Fiest, et al. 2014) Somboon et al., (2019) highlight how common insomnia is for individuals with epilepsy. Therefore item 3 on the PHQ (see appendix B) "*Trouble falling asleep, or sleeping too much*", may be measuring a symptom of epilepsy rather than depression. Indeed, item 4

("Feeling tired or having little energy") and item 7 ("Trouble concentrating on things, such as reading a newspaper or watching television") are similar to items on the LSSS (see appendix F; Item 4 "After my most severe seizures: I feel very confused" to "I do not feel confused at all"; Item 8, "After my most severe seizures: I always feel sleepy" to "I never feel sleepy"). If the high number of average yearly seizures (m= 120.62 per year) reported by patients with epilepsy is considered, it may be that elements of the PHQ-9 are also measuring symptom severity. It would also help to explain why there was a high degree of correlation between symptom severity, PHQ-9 and perceived stigma in the epilepsy group, but not in the NEAD group. A recent systemic review of depression screening tools for individuals with epilepsy by Gill et al. (2017) suggests that the Neurological Disorders Depression Inventory for Epilepsy (NDDI-E: Gilliam et al., 2006) may be a better tool for measuring depression in individuals with epilepsy than the PHQ-9. It should be noted, however, that the study by Rawlings, Brown, and Reuber (2017) used the NDDI-E and found a positive correlation between depression scores and perceived-stigma.

For future research, a longitudinal study, looking at stigma pre/post diagnosis might be helpful in understanding in causality in perceived stigma for NEAD, might help to address some of these limitations. A different comparison group for NEAD might also help to deconstruct perceived stigma in NEAD. It might also be beneficial for a different scale of stigma and depression to be used. The Stigma Scale (King et al., 2007), is a measure of stigma developed for use with people suffering from psychiatric and psychological difficulties. Therefore, it may be more suitable for NEAD and may be less influenced by the symptomatology of NEAD than the SSCI. The use of a different depression tool is more problematic, however, the use of NDDI-E may be indicated for the NEAD population, especially given the very high correlation of the PHQ-9 and the SSCI found in this study.

Conclusions and clinical implications

This study was a cross-sectional study, which investigated the relationship between self-efficacy, depression, anxiety, symptom severity, and perceived stigma in NEAD and epilepsy. The findings suggest that people with NEAD experience higher perceived stigma and depression than those with epilepsy, and that depression predicted stigma in both populations. Additional analysis found that symptom severity was not linked to stigma in NEAD but it was in epilepsy. Furthermore, that self-efficacy had a negative relationship with stigma in NEAD but not in epilepsy. This is an important finding for clinicians who work with NEAD, which suggests that depression might be a significant factor to target when planning therapeutic interventions. It would be beneficial for future research to consider longitudinal studies; charting perceived stigma pre- and post-diagnosis, as well as pre-and post-therapeutic intervention/treatment. Such studies would help develop a more comprehensive model of stigma in NEAD and how this may relate to future targeted interventions for this population.

References

Alonso, J., Buron, A., Rojas-Farreras, S., De Graaf, R., Haro, J. M., De Girolamo, G., ... & ESEMeD/MHEDEA 2000 Investigators. (2009). Perceived stigma among individuals with common mental disorders. *Journal of affective disorders, 118*, 180-186. https://doi.org/10.1016/j.jad.2009.02.006

Altın, M. (2018). Treatment of depression and anxiety disorders in patients with epilepsy. *Klinik Psikofarmakoloji Bulteni, 28,* 370-371. http://dx.doi.org/10.1136/jnnp.2004.060467

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Arlington, VA: American Psychiatric Publishing.

Arslan, G. (2018). Psychological maltreatment, social acceptance, social connectedness, and subjective well-being in adolescents. *Journal of Happiness Studies, 19,* 983-1001. https://doi.org/10.1007/s10902-017-9856-z

Bandura, A. (2010). Self-efficacy. *The Corsini encyclopedia of psychology, 1-3.* https://doi.org/10.1002/9780470479216.corpsy0836

Baskind, R., & Birbeck, G. L. (2005). Epilepsy-associated stigma in sub-Saharan Africa: the social landscape of a disease. *Epilepsy & Behavior, 7,* 68-73. https://doi.org/10.1016/j.yebeh.2005.04.009

Baldwin, K. M., Baldwin, J. R., & Ewald, T. (2006). The relationship among shame, guilt, and self-efficacy. *American Journal of Psychotherapy, 60,* 1-21. https://doi.org/10.1176/appi.psychotherapy.2006.60.1.1

Bansal, A., & Burton, C. (2019). Medically unexplained symptoms: What's in a name?. *British journal of General Practise*, 688. https://doi.org/10.3399/bjgp17X689473.

Benbadis, S. R. (2005). Psychogenic non-epileptic seizures. The treatment of epilepsy: principles and practice, 4, 623-630. https://doi.org/10.1016/j.yebeh.2005.04.009

Benbadis, S. R., & Hauser, W. A. (2000). An estimate of the prevalence of psychogenic non-epileptic seizures. *Seizure, 9*, 280-281. https://doi.org/10.1053/seiz.2000.0409

Benight, C. C., & Bandura, A. (2004). Social cognitive theory of posttraumatic recovery: The role of perceived self-efficacy. Behaviour research and therapy, 42(10), 1129-1148. https://doi.org/10.1016/j.brat.2003.08.008

Böhm, D., Stock Gissendanner, S., Bangemann, K., Snitjer, I., Werfel, T., Weyergraf, A., ... & Schmid-Ott, G. (2013). Perceived relationships between severity of psoriasis symptoms, gender, stigmatization and quality of life. *Journal of the European Academy of Dermatology and Venereology, 27*, 220-226. DOI: 10.1111/j.1468-3083.2012.04451.x

Bosco, F. A., Aguinis, H., Singh, K., Field, J. G., & Pierce, C. A. (2015). Correlational effect size benchmarks. *Journal of Applied Psychology, 100*, 431.

Brohan, E., Gauci, D., Sartorius, N., Thornicroft, G., & GAMIAN–Europe Study Group. (2011). Self-stigma, empowerment and perceived discrimination among people with bipolar disorder or depression in 13 European countries: The GAMIAN–Europe study. *Journal of affective disorders, 129*, 56-63. https://doi.org/10.1016/j.jad.2010.09.001

Brown, R. J. (2004). Psychological mechanisms of medically unexplained symptoms: an integrative conceptual model. *Psychological bulletin, 130*, 793. http://dx.doi.org/10.1037/0033-2909.130.5.793

Brown, R. J., & Reuber, M. (2016). Towards an integrative theory of psychogenic non-epileptic seizures (NEAD). Clinical psychology review, 47, 55-70. https://doi.org/10.1016/j.cpr.2016.06.003

Cataldo, J. K., Jahan, T. M., & Pongquan, V. L. (2012). Lung cancer stigma, depression, and quality of life among ever and never smokers. *European Journal of Oncology Nursing, 16,* 264-269. https://doi.org/10.1016/j.ejon.2011.06.008

Cataldo, J. K., & Brodsky, J. L. (2013). Lung cancer stigma, anxiety, depression and symptom severity. *Oncology, 85,* 33-40. doi: 10.1159/000350834

Claesson, K., Birgegard, A., & Sohlberg, S. (2007). Shame: Mechanisms of Activation and Consequences for Social Perception, Self-Image, and General Negative Emotion. *Journal of personality, 75,* 595-628.

Cooper, S., Campbell, G., Larance, B., Murnion, B., & Nielsen, S. (2018). Perceived stigma and social support in treatment for pharmaceutical opioid dependence. *Drug and alcohol review, 37,* 262-272. https://doi.org/10.1111/dar.12601

Cohen, J. (1992). Statistical power analysis. *Current directions in psychological science, 1,* 98-101. https://doi.org/10.1111/1467-8721.ep10768783

Connolly, J. (1989). Social self-efficacy in adolescence: Relations with self-concept, social adjustment, and mental health. *Canadian Journal of Behavioural Science/Revue canadienne des sciences du comportement, 21*, 258.

Corrigan, P. W. (2007). How clinical diagnosis might exacerbate the stigma of mental illness. *Social Work, 52,* 31-39. https://doi.org/10.1093/sw/52.1.31

Corrigan, P. W., Markowitz, F. E., & Watson, A. C. (2004). Structural levels of mental illness stigma and discrimination. *Schizophrenia bulletin, 30,* 481-491.

Corrigan, P. W., Watson, A. C., & Barr, L. (2006). The self–stigma of mental illness: Implications for self–esteem and self–efficacy. *Journal of social and clinical psychology, 25*, 875-884. doi: 10.1093/schbul/sbl076

Couture, S., & Penn, D. (2003). Interpersonal contact and the stigma of mental illness: A review of the literature. *Journal of mental health, 12*, 291-305. https://doi.org/10.1080/09638231000118276

Cull, C., & Goldstein, L. H. (Eds.). (2002). *The clinical psychologist's handbook of epilepsy: assessment and management.* Routledge.

DeWall, C. N., Twenge, J. M., Bushman, B., Im, C., & Williams, K. (2010). A little acceptance goes a long way: applying social impact theory to the rejection-aggression link. *Social Psychological and Personality Science, 1*, 168-174. https://doi.org/10.1177/1948550610361387

De Boer, H. M., Mula, M., & Sander, J. W. (2008). The global burden and stigma of epilepsy. *Epilepsy & behavior, 12*, 540-546. https://doi.org/10.1016/j.yebeh.2007.12.019

DeCou, C. R., Mahoney, C. T., Kaplan, S. P., & Lynch, S. M. (2019). Coping self-efficacy and trauma-related shame mediate the association between negative social reactions to sexual assault and PTSD symptoms. Psychological Trauma: Theory, Research, Practice, and Policy, 11(1), 51–54. https://doi.org/10.1037/tra0000379

DiIorio, C., Shafer, P. O., Letz, R., Henry, T., Schomer, D. L., Yeager, K., & Project EASE Study Group. (2003). The association of stigma with self-management and perceptions of health care among adults with epilepsy. *Epilepsy & Behavior, 4*, 259-267. https://doi.org/10.1016/S1525-5050(03)00103-3

de Souza, E. A. P., & Salgado, P. C. B. (2006). A psychosocial view of anxiety and depression in epilepsy. *Epilepsy & Behavior, 8*, 232-238. https://doi.org/10.1016/j.yebeh.2005.10.011

Faul, F., Erdfelder, E., Buchner, A., & Lang, A. G. (2009). Statistical power analyses using G* Power 3.1: Tests for correlation and regression analyses. *Behaviour research methods, 41*, 1149-1160. https://doi.org/10.3758/BF03193146

Fiest, K. M., Patten, S. B., Wiebe, S., Bulloch, A. G., Maxwell, C. J., & Jetté, N. (2014). Validating screening tools for depression in epilepsy. *Epilepsia, 55*, 1642-1650. https://doi.org/10.1111/epi.12754

Fisher, R. S., Acevedo, C., Arzimanoglou, A., Bogacz, A., Cross, J. H., Elger, C. E., ... & Hesdorffer, D. C. (2014). ILAE official report: a practical clinical definition of epilepsy. *Epilepsia, 55*, 475-482. 10.1111/epi.12550

Francis, P., & Baker, G. A. (1999). *Non-epileptic attack disorder (NEAD): a comprehensive review. Seizure, 8*, 53-61. https://doi.org/10.1053/seiz.1998.0246

Garnefski, N., van Rood, Y., De Roos, C., & Kraaij, V. (2017). Relationships between traumatic life events, cognitive emotion regulation strategies, and somatic complaints. *Journal of clinical psychology in medical settings, 24*, 144-151. https://doi.org/10.1007/s10880-017-9494-y

Gill, S. J., Lukmanji, S., Fiest, K. M., Patten, S. B., Wiebe, S., & Jetté, N. (2017). Depression screening tools in persons with epilepsy: a systematic review of validated tools. *Epilepsia, 58*, 695-705. 10.1111/epi.13651

Gilliam, F. G., Barry, J. J., Hermann, B. P., Meador, K. J., Vahle, V., & Kanner, A. M. (2006). Rapid detection of major depression in epilepsy: a multicentre study. *The Lancet Neurology, 5*, 399-405. https://doi.org/10.1016/S1474-4422(06)70415-X

Goffman, E. (2009). Stigma: *Notes on the management of spoiled identity.* Simon and Schuster.

Greco, M. (2012). The classification and nomenclature of 'medically unexplained symptoms': Conflict, performativity and critique. *Social Science & Medicine, 75,* 2362-2369. https://doi.org/10.1016/j.socscimed.2012.09.010

Green, B., Norman, P., & Reuber, M. (2017). Attachment style, relationship quality, and psychological distress in patients with psychogenic non-epileptic seizures versus epilepsy. *Epilepsy & Behavior, 66,* 120-126. doi: 10.1016/j.yebeh.2016.10.015

Griffiths, K. M., Christensen, H., & Jorm, A. F. (2008). Predictors of depression stigma. *BMC psychiatry, 8,* 25. https://doi.org/10.1186/1471-244X-8-25

Hing, N., Nuske, E., Gainsbury, S. M., & Russell, A. M. (2016). Perceived stigma and self-stigma of problem gambling: perspectives of people with gambling problems. *International Gambling Studies, 16,* 31-48. https://doi.org/10.1080/14459795.2015.1092566

Holmes, E., Bourke, S., & Plumpton, C. (2019). *Attitudes towards epilepsy in the UK population: Results from a 2018 national survey. Seizure,* 65, 12-19. https://doi.org/10.1016/j.seizure.2018.12.012

Hatzenbuehler, M. L., Phelan, J. C., & Link, B. G. (2013). *Stigma as a fundamental cause of population health inequalities. American journal of public health, 103,* 813-821. doi: 10.2105/AJPH.2012.301069

Hatzenbuehler, M. L., & Link, B. G. (2014). Introduction to the special issue on structural stigma and health. *Social Science & Medicine*. 103. doi: 10.1016/j.socscimed.2013.12.017

IBM Corp (2017). *IBM SPSS Statistics for Windows, Version 25.0*. Armonk, NY: IBM Corp.

Jacoby, A., & Austin, J. K. (2007). *Social stigma for adults and children with epilepsy. Epilepsia*, 48, 6-9. 10.1111/j.1528-1167.2007.01391.x

Jacoby, A., Snape, D., & Baker, G. A. (2005). Epilepsy and social identity: the stigma of a chronic neurological disorder. *The Lancet Neurology, 4,* 171-178. https://doi.org/10.1016/S1474-4422(05)01014-8

Joormann, J., & Gotlib, I. H. (2006). Is this happiness I see? Biases in the identification of emotional facial expressions in depression and social phobia. *Journal of abnormal psychology, 115*, 705. 10.1037/0021-843X.115.4.705

Kanter, J. W., Rusch, L. C., & Brondino, M. J. (2008). Depression self-stigma: a new measure and preliminary findings. *The Journal of nervous and mental disease, 196*, 663-670. doi: 10.1097/NMD.0b013e318183f8af

King, M., Dinos, S., Shaw, J., Watson, R., Stevens, S., Passetti, F., ... & Serfaty, M. (2007). The Stigma Scale: development of a standardised measure of the stigma of mental illness. *The British Journal of Psychiatry, 190,* 248-254. https://doi.org/10.1192/bjp.bp.106.024638

Kleim, B., Vauth, R., Adam, G., Stieglitz, R. D., Hayward, P., & Corrigan, P. (2008). Perceived stigma predicts low self-efficacy and poor coping in schizophrenia. *Journal of Mental Health, 17*, 482-491. https://doi.org/10.1080/09638230701506283

LaFrance, W. C., Baker, G. A., Duncan, R., Goldstein, L. H., & Reuber, M. (2013). Minimum requirements for the diagnosis of psychogenic nonepileptic seizures: a staged approach. *Epilepsia, 54*, 2005-2018. doi: 10.1111/epi.12356

LaFrance, W. C., & Syc, S. (2009). Depression and symptoms affect quality of life in psychogenic nonepileptic seizures. *Neurology, 73,* 366-371. doi: 10.1212/WNL.0b013e3181b04c83

LaFrance, W. C., Miller, I. W., Ryan, C. E., Blum, A. S., Solomon, D. A., Kelley, J. E., & Keitner, G. I. (2009). Cognitive behavioral therapy for psychogenic nonepileptic seizures. *Epilepsy & Behavior, 14,* 591-596. https://doi.org/10.1016/j.yebeh.2009.02.016

Landeen, J. L., Seeman, M. V., Goering, P., & Streiner, D. (2007). Schizophrenia: Effect of Perceived Stigma on Two Dimensions of Recovery. Clinical Schizophrenia & Related Psychoses, 1(1).

Leppänen, J. M. (2006). Emotional information processing in mood disorders: a review of behavioral and neuroimaging findings. Current opinion in psychiatry, 19(1), 34-39.

Lillis, J., Levin, M. E., & Hayes, S. C. (2011). Exploring the relationship between body mass index and health-related quality of life: A pilot study of the impact of weight self-stigma and experiential avoidance. *Journal of Health Psychology*, *16*, 722-727. https://doi.org/10.1177/1359105310388321

Link, B. G., & Phelan, J. C. (2001). Conceptualizing stigma. *Annual review of Sociology*, *27*, 363-385. https://doi.org/10.1146/annurev.soc.27.1.363

Link, B., Castille, D. M., & Stuber, J. (2008). Stigma and coercion in the context of outpatient treatment for people with mental illnesses. *Social science & medicine*, *67*, 409-419. https://doi.org/10.1016/j.socscimed.2008.03.015

Livingston, J. D., & Boyd, J. E. (2010). Correlates and consequences of internalized stigma for people living with mental illness: A systematic review and meta-analysis. *Social science & medicine*, *71*, 2150-2161. https://doi.org/10.1016/j.socscimed.2010.09.030

Looper, K. J., & Kirmayer, L. J. (2004). Perceived stigma in functional somatic syndromes and comparable medical conditions. *Journal of psychosomatic research*, *57*, 373-378. https://doi.org/10.1016/j.jpsychores.2004.03.005

Luszczynska, A., Benight, C. C., & Cieslak, R. (2009). Self-efficacy and health-related outcomes of collective trauma: A systematic review. European Psychologist, 14(1), 51-62.

Maddux, J. E. (2016). *Self-efficacy. In Interpersonal and intrapersonal expectancies (*pp. 41-46). Routledge.

Major, B., & O'brien, L. T. (2005). The social psychology of stigma. *Annu. Rev. Psychol., 56*, 393-421. 10.1146/annurev.psych.56.091103.070137

Manos, R. C., Rusch, L. C., Kanter, J. W., & Clifford, L. M. (2009). Depression self-stigma as a mediator of the relationship between depression severity and avoidance. *Journal of Social and Clinical Psychology, 28*, 1128-1143. doi: 10.1521/jscp.2009.28.9.1128

Marks EM, Hunter MS. (2015) Medically unexplained symptoms: an acceptable term? *British Journal of Pain. 9*. 109-14. doi: 10.1177/2049463714535372.

McLaughlin, D. P., Pachana, N. A., & Mcfarland, K. (2008). Stigma, seizure frequency and quality of life: the impact of epilepsy in late adulthood. *Seizure, 17*, 281-287. https://doi.org/10.1016/j.seizure.2007.09.001

Miller, C. T., & Kaiser, C. R. (2001). A theoretical perspective on coping with stigma. *Journal of social issues, 57*, 7 https://doi.org/10.1111/0022-4537.00202

Moskowitz, A., Dorahy, M. J., & Schäfer, I. (Eds.). (2019). *Psychosis, Trauma and Dissociation: Evolving Perspectives on Severe Psychopathology.* Wiley-Blackwell.3-92.

Micic, G., Richardson, C., Cain, N., Reynolds, C., Bartel, K., Maddock, B., & Gradisar, M. (2019). Readiness to change and commitment as predictors of therapy compliance in adolescents with Delayed Sleep-Wake Phase Disorder. *Sleep medicine, 55*, 48-55. https://doi.org/10.1016/j.sleep.2018.12.002

Monzoni, C. M., Duncan, R., Grünewald, R., & Reuber, M. (2011). Are there interactional reasons why doctors may find it hard to tell patients that their physical symptoms may have

emotional causes? A conversation analytic study in neurology outpatients. *Patient education and counseling, 85*, 189-200. https://doi.org/10.1016/j.pec.2011.07.014

Mula, M. (2017). Depression in epilepsy. *Current opinion in neurology, 30*, 180-186. 10.1097/WCO.0000000000000431

Müller, A., Znoj, H., & Moggi, F. (2019). How Are Self-Efficacy and Motivation Related to Drinking Five Years after Residential Treatment? A Longitudinal Multicenter Study. *European addiction research, 8*, 1-11. doi: 10.1159/000500520

National Institute for health and Care Excellence (2012), *Epilepsies: diagnosis and management*

Newton, C. R., & Garcia, H. H. (2012). Epilepsy in poor regions of the world. The Lancet, 380, 1193-1201. doi: 10.1016/S0140-6736(12)61381-6.

Oliveira, G. N. M. D., Araújo Filho, G. M. D., Kummer, A., Salgado, J. V., Portela, E. J., Sousa-Pereira, S. R., & Teixeira, A. L. (2011). Neurological Disorders Depression Inventory for Epilepsy (NDDI-E): Brazilian version of a screening instrument. *Journal of Epilepsy and Clinical Neurophysiology, 17*, 49-53. https://doi.org/10.1016/j.yebeh.2010.07.013

Oto, M., Conway, P., McGonigal, A., Russell, A. J., & Duncan, R. (2005). Gender differences in psychogenic non-epileptic seizures. *Seizure, 14*, 33-39. https://doi.org/10.1016/j.seizure.2004.02.008

Ottenbreit, N. D., & Dobson, K. S. (2004). Avoidance and depression: the construction of the Cognitive–Behavioral Avoidance Scale. *Behaviour research and therapy, 42*, 293-313. https://doi.org/10.1016/S0005-7967(03)00140-2

Pachankis, J. E., Hatzenbuehler, M. L., Hickson, F., Weatherburn, P., Berg, R. C., Marcus, U., & Schmidt, A. J. (2015). Hidden from health: structural stigma, sexual orientation concealment,

and HIV across 38 countries in the European MSM Internet Survey. *AIDS*. 29, 1239. doi: 10.1097/QAD.0000000000000724

Prince, P. N., & Prince, C. R. (2002). Perceived stigma and community integration among clients of assertive community treatment. *Psychiatric Rehabilitation Journal, 25*, 323. doi: 10.1037/h0095005

Plummer, F., Manea, L., Trepel, D., & McMillan, D. (2016). Screening for anxiety disorders with the GAD-7 and GAD-2: a systematic review and diagnostic metaanalysis. *General hospital psychiatry, 39*, 24-31. doi: 10.1016/j.genhosppsych.2015.11.005

Pryor, J. B., Reeder, G. D., Yeadon, C., & Hesson-McInnis, M. (2004). A dual-process model of reactions to perceived stigma. Journal of personality and social psychology, 87(4), 436.

Pyne, J. M., Kuc, E. J., Schroeder, P. J., Fortney, J. C., Edlund, M., & Sullivan, G. (2004). Relationship between perceived stigma and depression severity. *The Journal of nervous and mental disease, 192*, 278-283. doi: 10.1097/01.nmd.0000120886.39886.a3

Rawlings, G. H., Brown, I., & Reuber, M. (2017). Deconstructing stigma in psychogenic nonepileptic seizures: an exploratory study. *Epilepsy & Behavior, 74*, 167-172. https://doi.org/10.1016/j.yebeh.2017.06.014

Rawlings, G. H., & Reuber, M. (2016). What patients say about living with psychogenic nonepileptic seizures: a systematic synthesis of qualitative studies. *Seizure, 41*, 100-111. https://doi.org/10.1016/j.seizure.2016.07.014

Rawlings, G. H., & Reuber, M. (2018). Health care practitioners' perceptions of psychogenic nonepileptic seizures: a systematic review of qualitative and quantitative studies. *Epilepsia, 59*, 1109-1123. doi: 10.1111/epi.14189

Rathore, J. S., Jehi, L. E., Fan, Y., Patel, S. I., Foldvary-Schaefer, N., Ramirez, M. J., ... & Tesar, G. E. (2014). Validation of the Patient Health Questionnaire-9 (PHQ-9) for depression screening in adults with epilepsy. *Epilepsy & Behavior, 37*, 215-220. doi: 10.1016/j.yebeh.2014.06.030

Reuber, M., Kanner, A. M., & Schachter, S. (2008). Are non-epileptic seizures a manifestation of neurologic pathology? Controversies in epilepsy and behaviour.

Reuber, M., & Brown, R. J. (2017). Understanding psychogenic nonepileptic seizures—Phenomenology, semiology and the Integrative Cognitive Model. *Seizure, 44*, 199-205. https://doi.org/10.1016/j.seizure.2016.10.029

Reuber, M., Fernandez, G., Bauer, J., Helmstaedter, C., & Elger, C. E. (2002). Diagnostic delay in psychogenic nonepileptic seizures. *Neurology, 58*, 493-495. http://dx.doi.org/10.1212/WNL.58.3.493

Rosenfield, S. (1997). Labelling mental illness: The effects of received services and perceived stigma on life satisfaction. *American Sociological Review, 57*, 660-672.

Ross, C. E. (2017). *Social causes of psychological distress.* Routledge. UK: London

Rüsch, N., Angermeyer, M. C., & Corrigan, P. W. (2005). Mental illness stigma: Concepts, consequences, and initiatives to reduce stigma. *European psychiatry, 20*, 529-539. https://doi.org/10.1016/j.eurpsy.2005.04.004

Sampogna, F., Tabolli, S., & Abeni, D. (2012). Living with psoriasis: prevalence of shame, anger, worry, and problems in daily activities and social life. *Acta dermato-venereologica, 92*, 299-303. https://doi.org/10.2340/00015555-1273

Samuelson, K. W., Bartel, A., Valadez, R., & Jordan, J. T. (2017). PTSD symptoms and perception of cognitive problems: The roles of posttraumatic cognitions and trauma coping self-efficacy. Psychological Trauma: Theory, Research, Practice, and Policy, 9(5), 537.

Schomerus, G., Corrigan, P. W., Klauer, T., Kuwert, P., Freyberger, H. J., & Lucht, M. (2011). Self-stigma in alcohol dependence: consequences for drinking-refusal self-efficacy. *Drug and alcohol dependence, 114*, 12-17. https://doi.org/10.1016/j.drugalcdep.2010.08.013

Scott-Lennox, J., Bryant-Comstock, L., Lennox, R., & Baker, G. A. (2001). Reliability, validity and responsiveness of a revised scoring system for the Liverpool Seizure Severity Scale. Epilepsy research, 44(1), 53-63.

Somboon, T., Pascoe, M., Eippert, M., Bruton, M., Wang, L., Bena, J., & Foldvary-Schaefer, N. (2019). Insomnia in Epilepsy: Association Between Insomnia Symptom Severity and Disease-Related Characteristics. 56, 6-26

Shorvon, S., Perucca, E., & Engel Jr, J. (Eds.). (2015). *The treatment of epilepsy*. John Wiley & Sons.

Sickel, A. E., Seacat, J. D., & Nabors, N. A. (2014). Mental health stigma update: A review of consequences. *Advances in Mental Health, 12*, 202-215. https://doi.org/10.1080/18374905.2014.11081898

Sirven, J. I., Pedley, T. A., & Wilterdink, J. L. (2018). *Evaluation and management of drug-resistant epilepsy*. UpToDate, Waltham, MA. Accessed August, 13.

Smith, B. J. (2014). Closing the major gap in PNES research: finding a home for a borderland disorder. *Epilepsy currents, 14*, 63-67. doi: 10.5698/1535-7597-14.2.63

Sung, K. H. (2009). Self-perceived stigma, self-efficacy and quality of life in psychiatric outpatients. Journal of Korean Clinical Nursing Research, 15(2), 127-138.

Staring, A. B. P., Van der Gaag, M., Van den Berge, M., Duivenvoorden, H. J., & Mulder, C. L. (2009). Stigma moderates the associations of insight with depressed mood, low self-esteem,

and low quality of life in patients with schizophrenia spectrum disorders. 115, 363-369. doi: 10.1016/j.schres.2009.06.015

Szaflarski, J. P., & Szaflarski, M. (2004). Seizure disorders, depression, and health-related quality of life. *Epilepsy & Behavior, 5*, 50-57. https://doi.org/10.1016/j.yebeh.2003.10.015

Van Brakel, W. H. (2006). Measuring health-related stigma—a literature review. *Psychology, health & medicine, 11*, 307-334. https://doi.org/10.1080/13548500600595160

Taft, T. H., Keefer, L., Leonhard, C., & Nealon-Woods, M. (2009). Impact of perceived stigma on inflammatory bowel disease patient outcomes. *Inflammatory bowel diseases, 15*, 1224-1232. doi: 10.1002/ibd.20864

Taft, T. H., Keefer, L., Artz, C., Bratten, J., & Jones, M. P. (2011). Perceptions of illness stigma in patients with inflammatory bowel disease and irritable bowel syndrome. *Quality of Life Research, 20*, 1391. https://doi.org/10.1007/s11136-011-9883-x

Teachman, B. A., Wilson, J. G., & Komarovskaya, I. (2006). Implicit and explicit stigma of mental illness in diagnosed and healthy samples. *Journal of Social and Clinical Psychology, 25*, 75-95. https://doi.org/10.1521/jscp.2006.25.1.75

Trindade, I. A., Ferreira, C., & Pinto Gouveia, J. (2017). Chronic illness related shame: Development of a new scale and novel approach for IBD patients' depressive symptomatology. *Clinical psychology & psychotherapy, 24*, 255-263. https://doi.org/10.1002/cpp.2035

Wells, A., & Matthews, G. (1994). Attention and emotion: A clinical perspective. Hove, England: Erlbaum.

World Health Organization. (2018). *International classification of diseases for mortality and morbidity statistics (11th Revision).* Retrieved from https://icd.who.int/browse11/l-m/en

Yildirim, Z., Ertem, D. H., Dirican, A. C., & Baybas, S. (2019). Who is the bigger stigmatizor?: The loved one or the society. Epilepsy & Behavior, 96, 13-22. https://doi.org/10.1016/j.yebeh.2019.04.013

Appendix A – Information Sheet

Sheffield Teaching Hospitals NHS
NHS Foundation Trust
Excellence as standard

The University Of Sheffield.

PARTICIPANT INFORMATION SHEET – Version 1.2

Title of Project: The relationship between Stigma and Self-Efficacy in people with epilepsy or nonepileptic attack disorder

**Name of Researchers: Lewis Hanney,
Prof Markus Reuber and Dr. Andrew Thompson**

We would like to invite you to take part in a research study. It is important to understand the rationale for the research and what it will involve for you, before agreeing to participate in this study. Please read the following information carefully and feel free to contact the researchers if you have any questions or comments. It might also be useful to talk to others about the study. Thank you for reading this.

Background

People with epilepsy and non-epileptic attack disorder (NEAD) often experience significant stigma in their lives. Stigma can contribute to disability and is usually attributed to their condition. The degree to which people think that they can achieve their own goals has been shown to affect how well people manage with other conditions, and how badly stigmatised they feel. Changing these perceptions could help people to feel less stigmatised by their seizure disorder. We would like to investigate if what you think about yourself is an important factor in how stigmatised you feel.

This study is being carried out as part of a Doctor of Clinical Psychology (D. Clin. Psy.) research project based at the University of Sheffield.

What is the purpose of the study?

The purpose of this study is to investigate if people's thoughts about themselves affect their perception of stigma.

Sheffield Teaching Hospitals is the sponsor for this study based in the United Kingdom. We will be using information from you and your medical records in order to undertake this study and will act as the data controller for this study. This means that we are responsible for looking after your information and using it properly. Sheffield Teaching Hospitals will keep identifiable information about you for 10 years after the study has finished

Your rights to access, change or move your information are limited, as we need to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, we will keep the information about you that

we have already obtained. To safeguard your rights, we will use the minimum personally-identifiable information possible.

You can find out more about how we use your information by contacting Lewis Hanney (contact information provided below).

Why have I been asked to take part?
We are contacting people who have experienced seizures and who have been seeing a neurologist at the Royal Hallamshire Hospital in Sheffield. We are asking people with epileptic seizures as well as people with NEAD to take part in this study. We are not asking you to decide if you wish to participate in the study now. A member of the research team will approach you during your visit to the clinic and be available for any questions you may have.

Do I have to take part?
No. The study is completely voluntary and it is entirely up to you if you would like to participate.

Please feel free to contact a member of the research team if you have any questions or would like additional information. Your decision to participate or not will have no impact upon the care you receive at the seizure clinic.

What will happen to me if I take part? When you arrive for your appointment at the seizure clinic at the Royal Hallamshire Hospital, a member of the research team will approach you and you will have the opportunity to ask questions and discuss the research project. If you decide to participate, which you are not obliged to do, then you will be asked to complete a consent form, recording your agreement to take part. You will then be asked to complete a set of questionnaires, which should not take longer than 25 minutes.

What are the possible benefits of this study?
There is little current research on self-efficacy and stigma in people who experience seizures. This will help us develop our understanding of the role of stigma in people who experience seizures.

What are the possible risks of taking part in this study?
There are no significant risks associated with taking part in the study. Two of the questionnaires ask about your experience of symptoms of anxiety and depression. If the outcomes of these questionnaires raise any issues or concerns, then we will offer you appropriate support and provide you on the details of services that may also be able to offer you support. The researchers would also inform your clinician if you were likely to have anxiety or depression requiring treatment.

Will my taking part in this study be kept confidential?
All the information that is collected about you during this study will be kept strictly confidential. We will keep your personal details separate to your questionnaire responses. Some information will be kept in a locked cabinet in a secure location at the Academic Neurology Unit, University of Sheffield. This is because this is where the research team are based and is the most secure location. Only the research team will have access to your data. Personal details collected during the study will be destroyed once the study has finished. Anonymous study data will be kept for 10 years and then destroyed. We would only pass on clinically relevant findings (for example if we feel you may benefit from support related to the anxiety or depression questionnaires) to your consultant neurologist. We may also share information if there is a concern about risk to yourself or another person.

The research team will use your name, and contact details to contact you about the research study, and make sure that relevant information about the study is recorded for your care, and to oversee the quality of the study. Individuals from Sheffield Teaching Hospital and regulatory organisations may look at your medical and research records to check the accuracy of the research study. The research team will pass these details to Sheffield Teaching Hospital along with the information collected from you and/or your medical records. The only people in Sheffield Teaching Hospital who will have access to information that identifies you will be people who need to contact you to audit the data collection process. The people who analyse the information will not be able to identify you and will not be able to find out your name or contact details.

Sheffield Teaching Hospital will keep identifiable information about you from this study for 10 years after the study has finished.

Sheffield Teaching Hospital will collect information about you for this research study from your Medical records. This information will include your name and NHS number and health information, which is regarded as a special category of information. We will use this information to confirm your diagnosis.

What will happen to the results of the study?
The results of this study will contribute to a Doctor of Clinical Psychology (D. Clin. Psy.) thesis. We will also aim to publish the results of the study in a scientific journal. Any data presented will be anonymized and you will not be identifiable. Please let us know if you would like a summary of the results of the study once it is complete.

What if I change my mind?
You are under no obligation to participate in this study.

Research funding

This research project is funded by the University of Sheffield.

Who has reviewed this study?

NHS Ethics and The University of Sheffield

Who should I contact if I have a question or need more information?

Lewis Hanney
Clinical Psychology Unit
The University of Sheffield
Cathedral Court Floor F
1 Vicar Lane
Sheffield
S1 2LT
UK

Email: lhanney1@sheffeild.ac.uk

You can also contact with the research support officer: 0114 2226650, and Lewis will return your call

What if something goes wrong?
Please contact us should you have any concerns relating to this study and we will do our best to address your concerns.
If we are unable to be of help then you can make a complaint regarding the study, via Sheffield Patient Services Team (previously known as PALS) on 0114 2712400.

Alternatively you can contact the Academic Supervisor for the study:

Dr. Andrew Thompson

Clinical Psychology Unit
The University of Sheffield
Cathedral Court Floor F
1 Vicar Lane
Sheffield
S1 2LT
UK
a.r.thompson@sheffield.ac.uk

You can outline your concerns by filling out an anonymous online feedback form provided by Sheffield Teaching Hospitals NHS Foundation Trust at: https://www.sth.nhs.uk/patients/patient-experience/feedback/leave-feedback.

Organisations for further support

NHS Direct
Tel: 0845 46 47
Website: www.nhsdirect.nhs.uk

Mind, the mental health charity
Tel: 0300 123 3393
Website: www.mind.org.uk

Samaritans
Tel: 08457 90 90 90
Website: www.samaritans.org

Breathing Space
Tel: 0800 83 85 87
Website: www.breathingspacescotland.co.uk

Epilepsy Action
Tel: 0808 800 5050
Website: https://www.epilepsy.org.uk

Epilepsy Society
Tel: 01494 601 400
Website: http://www.epilepsysociety.org.uk

Appendix B – Consent forms

Appendix E: Consent Form (Online) –
Version 1.2 04/09/2018 IRAS : 237613 STH20255

Sheffield Teaching Hospitals NHS
NHS Foundation Trust
Excellence as standard

The University Of Sheffield.

CONSENT FORM – Online Participant Version 1.2

Title of Project: The relationship between Stigma and Self-Efficacy in people with epilepsy or nonepileptic attack disorder

Name of Researchers: Lewis Hanney, Prof. Markus Reuber, Dr. Andrew Thompson

	Please initial box
1. I confirm that I have read and understand the information sheet for the above study and have had the opportunity to ask questions.	☐
2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without consequence.	☐
3. I understand that relevant sections of my medical notes and data collected during the study may be looked at by members of the research team, regulatory authorities or the Research Department at Sheffield Teaching Hospitals NHS Foundation, where it is related to my participation in this research. I give permission for these individuals to have access to my records and for my diagnosis to be checked from my records	☐
4. I am happy for my GP to share information about my diagnosis with the research so that we can confirm the diagnosis for this research study	☐
5. I agree that the research team can inform my General Practitioner about evidence of likely anxiety or depression	☐
6. I agree to take part in the above study and understand that the data will be used as part of a Doctor of Clinical Psychology (DClinPsy) degree thesis and may be used for publication.	☐

7. I agree for the researchers to return a protected copy of the countersigned consent form by email. If I do not consent then a copy of this form will be sent via post. ☐

   Please type your name in the signature box below in lieu of a signature to allow us to take online consent

Name	Date of birth	Signature

GP details

GP: _____

Practice: _____

_____

_____

Name of person receiving consent	Date	Signature

I

Appendix C – Ethics approval

Health Research Authority

Yorkshire & The Humber - South Yorkshire Research Ethics Committee

NHSBT Newcastle Blood Donor Centre
Holland Drive
Newcastle upon Tyne
NE2 4NQ

Telephone: 0207 1048091

Please note: This is the favourable opinion of the REC only and does not allow you to start your study at NHS sites in England until you receive HRA Approval

15 October 2018

Professor Markus Reuber
Professor of Clinical Neurology
University of Sheffield
Academic Neurology
University of Sheffield Royal Hallamshire Hospital Glossop Road
Sheffield
S10 2 JF

Dear Professor Reuber

Study title:	**The relationship between Stigma and Self-Efficacy in individuals with epilepsy or nonepileptic attack disorder**
REC reference:	**18/YH/0283**
IRAS project ID:	**237613**

Thank you for your letter of 9 October 2018, responding to the Committee's request for further information on the above research [and submitting revised documentation].

The further information has been considered on behalf of the Committee by the Chair and two named members.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this opinion letter. Should you wish to provide a substitute contact point, require further

information, or wish to make a request to postpone publication, please contact hra.studyregistration@nhs.net outlining the reasons for your request.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a **favourable ethical opinion** for the above research on the basis described in the application form, protocol and supporting documentation [as revised], subject to the conditions specified below.

Conditions of the favourable opinion

The REC favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements. Each NHS organisation must confirm through the signing of agreements and/or other documents that it has given permission for the research to proceed (except where explicitly specified otherwise).

Guidance on applying for HRA and HCRW Approval (England and Wales)/ NHS permission for research is available in the Integrated Research Application System, at www.hra.nhs.uk or at http://www.rdforum.nhs.uk.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of management permissions from host organisations

Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database within 6 weeks of recruitment of the first participant (for medical device studies, within the timeline determined by the current registration and publication trees).

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g. when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non-clinical trials this is not currently mandatory.

If a sponsor wishes to request a deferral for study registration within the required timeframe, they should contact hra.studyregistration@nhs.net. The expectation is that all clinical trials will be registered, however, in exceptional circumstances non registration may be permissible with prior agreement from the HRA. Guidance on where to register is provided on the HRA website.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Ethical review of research sites

NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

Document	*Version*	*Date*
Copies of advertisement materials for research participants	1.2	11 September 2018
GP/consultant information sheets or letters [GP Letter - High depression/anxiety score]	1.2	15 March 2018
GP/consultant information sheets or letters [GP Letter - Online Recruitment]	1.2	04 September 2018
IRAS Application Form [IRAS_Form_09102018]		09 October 2018
IRAS Checklist XML [Checklist_09102018]		09 October 2018
Letter from statistician		23 January 2018
Letters of invitation to participant	1	13 April 2018
Other [Appendix J participant email form]	1	10 August 2018
Other [Consent form Clinic]	1.2	04 September 2018
Other [HRA Responce Letter]	1	14 September 2018
Participant consent form	1.2	
Participant information sheet (PIS)	1.2	11 September 2018
Referee's report or other scientific critique report		23 January 2018
Research protocol or project proposal [Research protocol]	1	13 March 2018
Research protocol or project proposal [Research protocol]	1.2	04 September 2018
Summary CV for Chief Investigator (CI) [CV for Iras]		14 May 2018
Summary CV for student		
Summary CV for student [LH]		
Summary CV for supervisor (student research)		
Summary CV for supervisor (student research) [AT]		
Summary, synopsis or diagram (flowchart) of protocol in non technical language	1.2	04 September 2018

Validated questionnaire [Patient Health Questionairre-9]		
Validated questionnaire [Generalised Anxiety Disorder-7]		
Validated questionnaire [Stigma Scale for Chronic Illnesses 8]		
Validated questionnaire [Liverpool Seizure Severity Scale]		
Validated questionnaire [The General Self-Efficacy Scale]		

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Reporting requirements

The attached document *"After ethical review – guidance for researchers"* gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The HRA website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website:
http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/

HRA Training

We are pleased to welcome researchers and R&D staff at our training days – see details at
http://www.hra.nhs.uk/hra-training/

18/YH/0283	**Please quote this number on all correspondence**

With the Committee's best wishes for the success of this project.

Yours sincerely

pp

Dr Ian Woollands
Chair

Email: nrescommittee.yorkandhumber-southyorks@nhs.net

Enclosures: "After ethical review – guidance for researchers"

Copy to: Ms Samantha Walmsley Walmsley, Sheffield Teaching Hospitals NHS Foundation Trust

Ymchwil Iechyd a Gofal Cymru
Health and Care Research Wales

NHS
Health Research Authority

Professor Markus Reuber
Professor of Clinical Neurology
University of Sheffield
Academic Neurology
Glossop Road
Sheffield
S10 2JF

Email: hra.approval@nhs.net
Research-permissions@wales.nhs.uk

15 October 2018

Dear Professor Reuber

HRA and Health and Care Research Wales (HCRW) Approval Letter

Study title:	**The relationship between Stigma and Self-Efficacy in individuals with epilepsy or nonepileptic attack disorder**
IRAS project ID:	**237613**
REC reference:	**18/YH/0283**
Sponsor	**Sheffield Teaching Hospitals NHS FT**

I am pleased to confirm that HRA and Health and Care Research Wales (HCRW) Approval has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further relating to this application.

How should I continue to work with participating NHS organisations in England and Wales?
You should now provide a copy of this letter to all participating NHS organisations in England and Wales, as well as any documentation that has been updated as a result of the assessment.

This is a single site study sponsored by the site. The sponsor R&D office will confirm to you when the study can start following issue of HRA and HCRW Approval.

It is important that you involve both the research management function (e.g. R&D office) supporting each organisation and the local research team (where there is one) in setting up your study. Contact details of the research management function for each organisation can be accessed here.

How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland?
HRA and HCRW Approval does not apply to NHS/HSC organisations within the devolved administrations of Northern Ireland and Scotland.

IRAS project ID	237613

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance report (including this letter) has been sent to the coordinating centre of each participating nation. You should work with the relevant national coordinating functions to ensure any nation specific checks are complete, and with each site so that they are able to give management permission for the study to begin.

Please see IRAS Help for information on working with NHS/HSC organisations in Northern Ireland and Scotland.

How should I work with participating non-NHS organisations?
HRA and HCRW Approval does not apply to non-NHS organisations. You should work with your non-NHS organisations to obtain local agreement in accordance with their procedures.

What are my notification responsibilities during the study?
The document "*After Ethical Review – guidance for sponsors and investigators*", issued with your REC favourable opinion, gives detailed guidance on reporting expectations for studies, including:

- Registration of research
- Notifying amendments
- Notifying the end of the study

The HRA website also provides guidance on these topics, and is updated in the light of changes in reporting expectations or procedures.

I am a participating NHS organisation in England or Wales. What should I do once I receive this letter?
You should work with the applicant and sponsor to complete any outstanding arrangements so you are able to confirm capacity and capability in line with the information provided in this letter.

The sponsor contact for this application is as follows:

Name: Mrs Samantha Walmsley
Tel: 01142265937
Email: samantha.walmsley@sth.nhs.uk

Who should I contact for further information?
Please do not hesitate to contact me for assistance with this application. My contact details are below.

Your IRAS project ID is **237613**. Please quote this on all correspondence.

Yours sincerely

Aliki Sifostratoudaki
Assessor

Email: hra.approval@nhs.net

Appendix D - Stigma Scale for Chronic Illness (SSCI-8: Molina, Choi, Cella & Rao, 2013)

Stigma Scale for Chronic Illnesses 8

	1= Never 2 = Rarely 3 = Sometimes 4 = Often 5 = Always				
Because of my illness, some people seemed uncomfortable with me.	1	2	3	4	5
Because of my illness, some people avoided me.	1	2	3	4	5
Because of my illness, I felt left out of things.	1	2	3	4	5
Because of my illness, people were unkind to me.	1	2	3	4	5
Because of my illness, people avoided looking at me.	1	2	3	4	5
I felt embarrassed about my illness.	1	2	3	4	5
I felt embarrassed because of my physical limitations.	1	2	3	4	5
Some people acted as though it was my fault I have this illness.	1	2	3	4	5

Appendix E: The General Self-Efficacy Scale (GSE: Schwarzer & Jerusalem, 1995)

The General Self-Efficacy Scale (GSE)

1 = Not at all true 2 = Hardly true 3 = Moderately true 4 = Exactly true

1	I can always manage to solve difficult problems if I try hard enough.	1	2	3	4
2	If someone opposes me, I can find the means and ways to get what I want.	1	2	3	4
3	It is easy for me to stick to my aims and accomplish my goals.	1	2	3	4
4	I am confident that I could deal efficiently with unexpected events.	1	2	3	4
5	Thanks to my resourcefulness, I know how to handle unforeseen situations.	1	2	3	4
6	I can solve most problems if I invest the necessary effort.	1	2	3	4
7	I can remain calm when facing difficulties because I can rely on my coping abilities.	1	2	3	4
8	When I am confronted with a problem, I can usually find several solutions.	1	2	3	4
9	If I am in trouble, I can usually think of a solution.	1	2	3	4
10	I can usually handle whatever comes my way.	1	2	3	4

Appendix F: Generalised Anxiety Disorder-7 (GAD-7: Lowe et al., 2008).

Generalised Anxiety Disorder-7 (GAD-7)

Over the last 2 weeks, how often have you been bothered by the following problems?			
0 Not at all	1 Several days	2 Over half the days	3 Nearly every day
GAD-7			
1) Feeling nervous, anxious, or on edge			0 1 2 3
2) Not being able to stop or control worrying			0 1 2 3
3) Worrying too much about different things			0 1 2 3
4) Trouble relaxing			0 1 2 3
5) Being so restless that it's hard to sit still			0 1 2 3
6) Becoming easily annoyed or irritable			0 1 2 3
7) Feeling afraid as if something awful might happen			0 1 2 3

Appendix G: Patient Health-Questionairre-9 (PHQ-9: Kroenke, Spitzer, & Williams, 2002).

Patient Health Questionairre-9 (PHQ-9)

Over the last 2 weeks, how often have you been bothered by the following problems?			
0 Not at all	1 Several days	2 Over half the days	3 Nearly every day
PHQ-9			0 1 2 3
1) Little interest or pleasure in doing things			0 1 2 3
2) Feeling down, depressed or hopeless			0 1 2 3
3) Trouble falling asleep, or sleeping too much			0 1 2 3
4) Feeling tired or having little energy			0 1 2 3
5) Poor appetite or overeating			0 1 2 3
6) Feeling bad about yourself- or that you are a failure or have let your family down			0 1 2 3
7) Trouble concentrating on things, such as reading the newspaper or watching television			0 1 2 3
8) Moving or speaking so slowly that other people could have noticed. Or the opposite- being so fidgety and restless that you have been moving around a lot more than usual.			0 1 2 3
9) Thoughts that you would be better off dead or hurting yourself in some way.			0 1 2 3

Appendix H: Liverpool Seizure Severity Scale – Revised (LSSS-3: Scott-Lennox, Bryant-Comstock, Lennox, & Barker, 2001)

Liverpool Seizure Severity Scale LSSS

So we can better understand the frequency and severity of your seizures, please complete the following questionnaire about your seizures.

How often have you experienced seizures in the **past year**?

Please give an estimate of the number of seizures you usually had in a day, a week, a month or a year (please use the time period for which it is easiest for you to make an estimate, for example, '1 seizure per week' or '10 seizures per month'). Please enter '0' if you have not experienced any seizures in the past year.

_____ seizures per _____ (day/week/month/year)

How many seizures have you had in the **last 4 weeks**?

Please enter '0' if you have not experienced seizures in the last 4 weeks. If you cannot remember the exact number of seizures you've experienced, please estimate based on the number you usually had during a single day or week.

_____ seizures

Please complete the following questions thinking about the most severe seizure you experienced during the past 4 weeks. (This may be different for each individual, but is based on your most severe seizures over the past 4 weeks).

Please answer each question based on the most severe seizure you have experienced in the past 4 weeks. Circle only one answer for each question.

1. I feel that my most severe seizures have mostly been:

Very severe 0
Severe 1
Mild 2
Very mild 3

---

2. Most commonly when I blank out/lose consciousness:

I blank out for less than 1 minute 1
I blank out for between 1 and 2 minutes 2
I blank out for between 3 and 5 minutes 3
I blank out for more than 5 minutes 4
I never blank out lose consciousness 0

---

3. When I have my most severe seizures, I smack my lips, fidget, or behave in an unusual way:

Always 0

Usually 1

Sometimes 2

Never 3

---

4. After my most severe seizures:

I feel very confused 0

I feel fairly confused 1

I feel slightly confused 2

I do not feel confused at all 3

---

5. After my most severe seizures my confusion lasts for:

Less than 1 minute 1

Between 1 and 5 minutes 2

Between 6 minutes and 1 hour 3

1 to 2 hours 4

More than 2 hours 5

I never feel confused 0

---

6. When I have my most severe seizures:

I always fall to the ground 0

I usually fall to the ground 1

I sometimes fall to the ground 2

I never fall to the ground 3

---

7. After my most severe seizures:

I always have a headache 0

I usually have a headache 1

I sometimes have a headache 2

I never have a headache 3

---

8. After my most severe seizures:

I always feel sleepy 0

I usually feel sleepy 1

I sometimes feel sleepy 2

I never feel sleepy 3

---

9. After my most severe seizures:

I always find that I have wet myself 0

I usually find that I have wet myself 1

I sometimes find that I have wet myself 2

I never find that I have wet myself 3

---

10. After my most severe seizures:

I always find that I have bitten my tongue 0

I usually find that I have bitten my tongue 1

I sometimes find that I have bitten my tongue 2

I never find that I have bitten my tongue 3

---

11. After my most severe seizures:

I always find that I have injured myself (other than biting my tongue)..... 0

I usually find that I have injured myself (other than biting my tongue)..... 1

I sometimes find that I have injured myself (other than biting my tongue)..... 2

I never find that I have injured myself (other than biting my tongue)..... 3

---

12. After my most severe seizures I can usually return to what I am doing in:

Less than 1 minute 0

Between 1 and 5 minutes 1

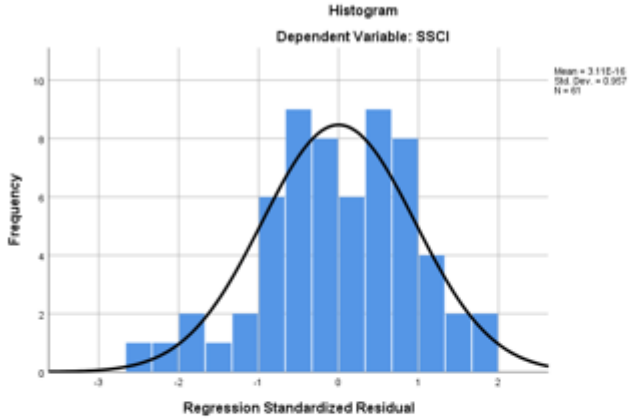
Between 6 minutes and 1 hour 2

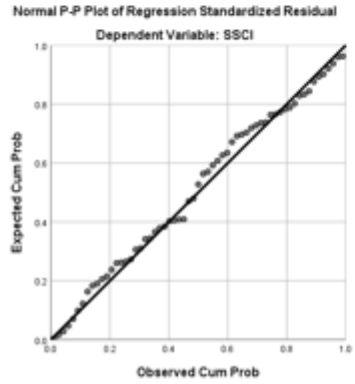
1 to 2 hours 3

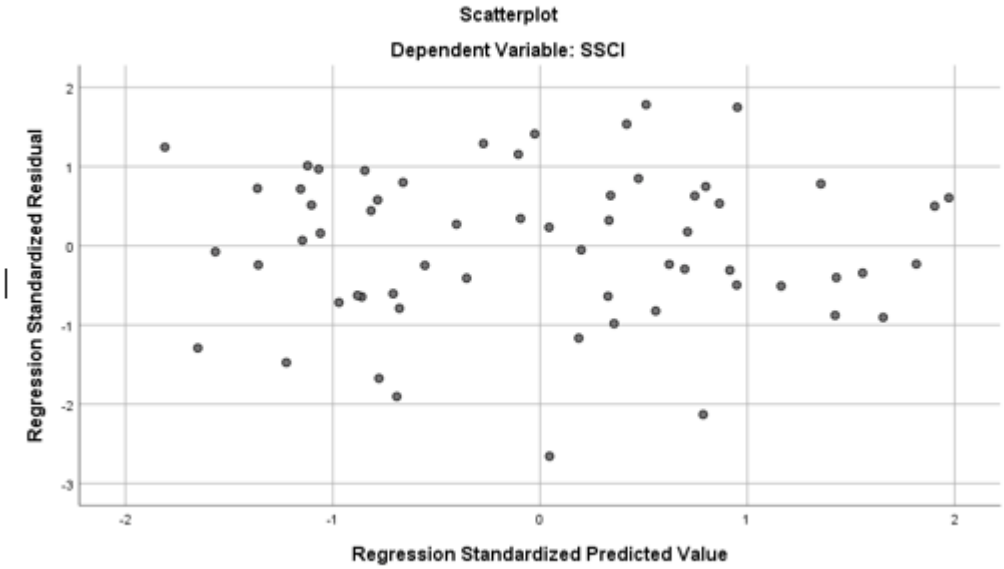
More than 2 hours 4

Please check that you have answered **all** the questions.

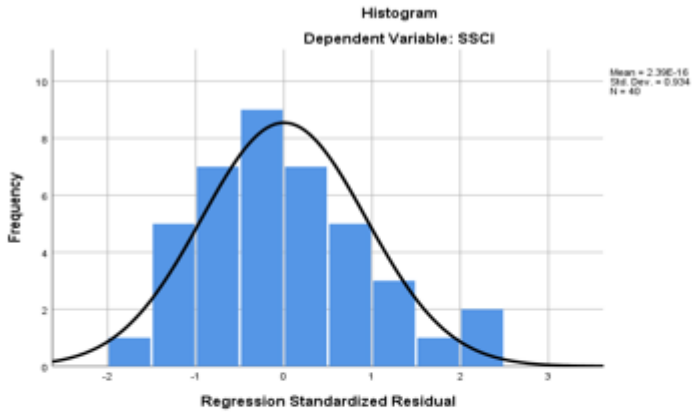
Appendix I: histogram of standardised residuals normal P-P plot of standardised residuals and scatterplot of standardised residuals for NEAD

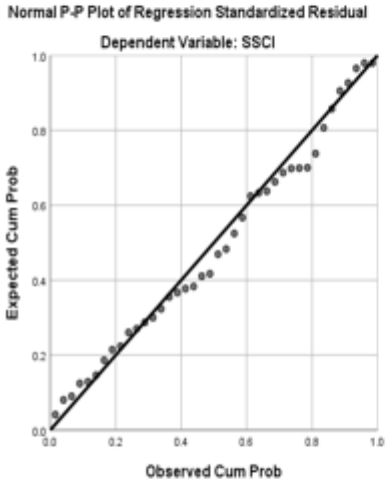


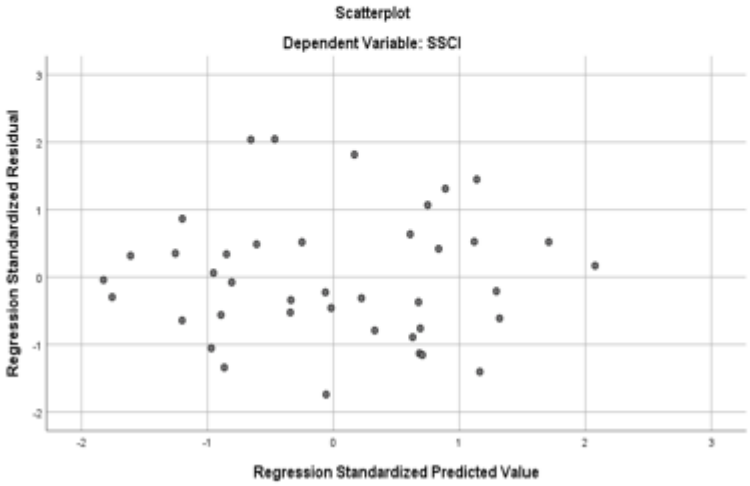




Appendix J: histogram of standardised residuals normal P-P plot of standardised residuals and scatterplot of standardised residuals for Epilepsy







Appendix K

An analysis of standard residuals was carried out, which indicated that the data contained no outlier (NEAD: Std. Residual Min = -2.66, Std. Residual Max = 1.78; Epilepsy: Std. Residual Min = -1.74, Std. Residual Max = 2.046).

The data met the assumption of independent errors (NEAD: Durbin-Watson value = 1.7; epilepsy: Durbin-Watson value = 1.44).

To check if the data met the assumption of collinearity, test indicated that multi-collinearity was not a concern (NEAD: PHQ9, Tolerance =.28, VIF = 3.54; GAD-7, Tolerance = .30, VIF = 3.33; GSE: Tolerance = .66, VIF= 1.5; LSSS = .94, VIF = 1.07, No. seizures per year, Tolerance = .95, VIF= 1.05; Epilepsy: PHQ9, Tolerance =.57, VIF = 1.76; GAD-7, Tolerance = .55, VIF = 1.81; GSE: Tolerance = .66, VIF= 1.5; LSSS = .87, VIF = 1.15, No. seizures per year, Tolerance = .98, VIF= 1.01).

The histogram of standardised residuals for both NEAD (appendix I) and epilepsy (appendix J) indicated that the data had approximately normally distributed errors The normal P-P plot of standardised residuals for NEAD (appendix I) and epilepsy (appendix J) showed points that were clustered closely around the line. The scatterplot of standardised residuals for NEAD (appendix I) and epilepsy (appendix J) indicated that the data met the assumptions of homogeneity of variance and linearity.

The assumption of non-zero variances was also met (NEAD: PHQ-9, variance = 62.54; GAD-7, variance = 43.7; GSE, variance = 44.82; SCCI-8, variance = 63.02; LSSS=71.07; Number of reported seizures per year, variance = 518091.65; Epilepsy: PHQ-9, variance = 42.1; GAD-7, variance = 47.11; GSE, variance = 73.01; SCCI-8, variance = 71.5; LSSS=71.5; Number of reported seizures per year, variance = 90454.43).